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Chapter 1

General introduction

Children usually have acute, short-term illnesses such as an upper respiratory tract infection or gastrointestinal illness with vomiting and diarrhea, or injury-related problems. Some children develop a chronic illness that lasts for years or even lifelong, because of genetic conditions, environmental factors, or a combination of both. Type 1 diabetes is among the common chronic conditions in children. There are two main types of diabetes: type 1 diabetes and type 2 diabetes. Type 1 diabetes typically presents in childhood or early adult life. Type 2 diabetes is often diagnosed after the age of 50 years. The present thesis focuses on type 1 diabetes mellitus (T1DM), the most common form in children. In recent years, the number of young children with T1DM is increasing worldwide [1-4]. Although the medical knowledge of T1DM has improved enormously over the last years, the psychological aspects, especially in young children and their parents, has received much less attention. Therefore, the present thesis focusses on the psychological aspects of young children (0-7 years) with T1DM and their parents.

The history of type 1 diabetes

As early as 1500 BC, diabetes symptoms were mentioned in ancient Egypt and India [5]. In the 2nd century AD, Aretaeus of Cappadocia was probably the first to describe 'diabetes', which refers to the Greek word διαβαινειν "to pass through" emphasizing the incessant flow of urine in these patients [6]. Recommendations for treatment of diabetes consisted of herbal drinks, drinking wine or milk, horseback riding, letting blood, using opium or eating as much as possible to stop losing weight [5, 6]. In the 17th and 18th centuries, it was found that the urine of these patients tasted sweet. Therefore, the adjective 'mellitus' was added, which refers to the Latin word for honey. Furthermore, Matthew Dobson discovered in 1776 that not only the urine of these patients tasted sweet, but that the blood of these patients also contained lots of sugar [5]. Soon thereafter, John Rollo described in 1797 that the consumption of food influenced people's blood sugar level, which was further clarified by Claude Bernard around 1847, who discovered that glucose was stored in the liver [5]. A few years later, in 1889, Oskar Minkowski and Josef von Mering discovered that the pancreas was involved in this mysterious disease called diabetes mellitus, by performing autopsy on a dog. Some particular cells in the pancreas, the *islands of Langerhans* (discovered by Paul Langerhans in 1869) were found responsible for making a secretion by Gustave Laguesse in 1893 [5]. This secretion, with which the body can process blood glucose, was called insulin (Latin for island) by Jean de Meyer in 1909 [5]. Then, it took about 20 years to be able to treat the first patients with insulin in 1922 (see Figure 1). This great discovery that has saved and prolonged so many lives was done by Banting, his assistant Best and MacLeod.

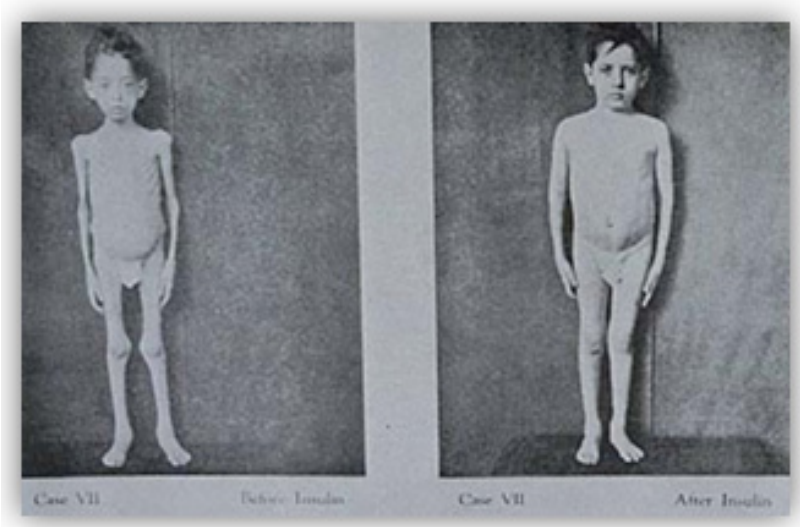

Figure 1: A patient treated with insulin: before insulin treatment (left) and after (right) [7].

However, "*the discovery of insulin was only the beginning, diabetes was a far more complicated disease than anyone had realized*" (quote from The Discovery of Insulin by Michael Bliss 1982 [8]).

What is type 1 diabetes?

The causes of T1DM still remain unknown, but it is assumed that genetic (a predisposition for developing T1DM), environmental (seasonality of onset and birth, nutrition) and autoimmune (viruses) factors are important factors in developing T1DM [9]. It is also clear that T1DM is an autoimmune disease, in which the insulin producing β-cells in most of the islets of Langerhans (in the pancreas) are destroyed or inactivated [10, 11]. Therefore, the body is no longer able producing insulin to transport glucose into cells for energy. This deficiency of insulin results in high blood glucose levels, which expresses itself by polyuria, polydipsia, weight loss, fatigue and could eventually result in ketoacidosis, stupor, coma and death [12, 13]. Patients with T1DM are fully dependent on insulin therapy for their survival. At first, after the famous discovery of insulin therapy by Frederick Banting and Charles Best in 1921, animal insulin was used which caused allergic reactions in some users. In the late 70's the first synthetically

manufactured insulin was produced, which was a much better option than animal insulin, because the synthetically manufactured insulin caused fewer side effects and was better tolerated by patients [5]. Nowadays, insulin is administered through multiple daily insulin injections or an insulin pump to achieve a 'near-normal' blood glucose level. By using an insulin pump, children with T1DM do not have to be injected several times a day, only once per 3 days, and therefore they are often less hindered to live a 'normal' life. Nevertheless, patients with T1DM have a complex and demanding treatment regimen, consisting of checking the blood glucose level several times a day, regulating food intake, administering the correct amount of insulin, and guarding these parameters in conjunction with the level of physical activity [14]. On top of this, factors such as stress, warm weather, taking different types of food, or fever can all impact glucose metabolism and glycemic control [5].

The incidence of type 1 diabetes is increasing, especially in young children

T1DM is one of the most common endocrine and metabolic conditions in pediatrics [14]. An increasing number of children is diagnosed with T1DM [1], with an annual increase of 3-5% [5], particularly in the youngest age group [1-3]. In Europe and North America about 1 in every 300 children will develop diabetes before they are 20 years old [5]. T1DM is more common in northern countries like Scandinavia [1]. In the Netherlands, T1DM is present in approximately 6000 children aged 0-18 years [15] and the incidence is also increasing, especially in the youngest age group (0-4 years) [4].

Glycosylated hemoglobin level is an important factor in type 1 diabetes

An important factor in the treatment of T1DM is the glycosylated hemoglobin level, or HbA_{1c} level. The HbA_{1c} level is measured once every 2-3 months and gives an impression of the average blood glucose level over a six to twelve week period [16]. Results of The Diabetes Control and Complications Trial [17] have convincingly shown that keeping HbA_{1c} levels close to normal levels avoids or delays the onset of long-term complications of diabetes, such as neuropathy, retinopathy, nephropathy and severe long-term health consequences as renal failure, heart disease, and blindness. In addition to these long-term complications, research has also shown that suboptimal HbA_{1c} levels are associated with short-term complications like child behavior problems [18-20], and a lower quality of life [21-23]. An important disadvantage of tight glycemic control is that there is an increased risk for hypoglycemia. Patients might experience a hypoglycemic event in which their blood glucose level is too low by injecting too much insulin, eating not enough food or exercising without extra food [14]. When experiencing a mild hypoglycemic event, children with T1DM

may feel a bit shaky, weak, nervous, and sweaty, have a headache, get blurred vision and hunger [14]. Untreated, a severe hypoglycemic event might result in unconsciousness, seizures, coma and eventually death [24]. As a result, parents of children with T1DM are fearful for hypoglycemia [25]. To avoid the long-term complications, parents need to keep their child's HbA_{1c} level close to the recommended level of 7.5% or 58 mmol/mol [26]. However, with lower HbA_{1c} levels, the risk of a hypoglycemic event increases [26]. Despite all efforts, the majority of children with T1DM still do not reach this optimal HbA_{1c} level [27, 28]. Reaching an optimal HbA_{1c} level, therefore, is needed to avoid medical risks like hypoglycemic events [26] and the onset of long-term complications [26] and is therefore seen as an important factor in the treatment of T1DM.

Quality of life is an important psychological outcome factor in type 1 diabetes

Because the treatment of T1DM is intensive and demanding, this could impair the quality of life (QoL) of these children. Therefore, improving or optimizing the QoL of these children is another important child outcome factor in the treatment of T1DM. Definitions of QoL vary considerably, although there is consensus that QoL should be regarded as a multidimensional construct which describes the physical, mental and social functioning of a person [29, 30].

Literature examining the QoL of children with T1DM report conflicting results as some studies report an impairment in QoL compared to healthy peers (e.g. [31, 32]), whereas other studies report the same level of QoL or that children with T1DM even show adaptive outcomes (e.g. [33, 34]). A possible explanation for impaired QoL could be the intensive treatment demands and possible hypoglycemic events. On the other hand, because of the substantial improvements in treatment regimen in the last decade [35, 36] it is also conceivable that, nowadays, it is easier to handle the demands of T1DM and therefore it could be that the QoL of children with T1DM is not impaired.

However, a systematic overview that summarizes the literature that has focused on the QoL of children with T1DM is lacking. Therefore, we have conducted such a systematic review, and we included case-control studies that compared the generic QoL of children and adolescents with T1DM with healthy controls. Furthermore, we also examined whether QoL differs between boys and girls with T1DM, whether QoL of children with T1DM differs across different developmental stages, and which diabetes-specific QoL domains are most affected in children and adolescents with T1DM (see **Chapter 2**).

The quality of parent-child interaction might be affected in families with a young child with type 1 diabetes

Looking at Erickson's early stages of development [37] (see Table 1), one can see that the first years are crucial in child development. Parents of children with T1DM often have to negotiate with their children when they have to check their blood glucose level, administer insulin or when they try to make sure that their child will eat the correct amount of carbohydrates. The latter is becoming less of a burden for parents as in modern, more flexible diabetes treatments, food guides insulin and not the other way around. If parents try to 'trick' their child, for example by administering insulin through an injection unexpectedly, this could lead to mistrust. Also, when parents do not respect the autonomy of the child at a later age, this could lead to doubt and shame in the child. Furthermore, when children are not able to cope with the challenges accompanied with T1DM and do not get increased responsibilities, this might lead to frustration and/or guilt. Also, if the children are not able to engage in social challenges due to their T1DM (for example, being ashamed for having T1DM or due to severe worries about hypoglycemic events), this might lead to feelings of inferiority and incompetence.

Table 1: Erickson's early stages of development [37]

Age	Children	Failure may lead to:
0-1 years	Develop a sense of trust in their caregivers, their selves and their environment	Mistrusting both themselves and others
1-3 years	Develop a sense of autonomy and independence from their caregivers	Shame and doubt
3-6 years	Try to cope with challenges and an increasing responsibility	Guilt and aggression
6+ years	Try to master intellectual and social challenges	Feelings of inferiority and incompetence

The parents need or wish to adhere to the recommended treatment regimen may interfere with normal and age appropriate behaviors of the children, which might be a challenge for these parents. One can imagine that when children begin to refuse food, display oppositional behavior or seeking independence (all normal toddler and pre-school age behaviors), this could interfere considerably with the ability of the parents to adhere to all the treatment demands [38]. Managing a 'picky eater', setting limits and coping with children who show a lack of cooperation with the treatment regimen [39], may affect family functioning and parent-child interactions [40-42]. The quality of the parent-child interaction in families with a young child with T1DM might be impaired [43]. Therefore, is it highly important for health care providers to be able to examine the parent-child interaction in families with a young child with

T1DM to see whether there is room for improvement.

However, there are only a small number of studies that have examined the quality of parent-child interaction in families with a child with T1DM; most studies have been conducted with older children (> 8 years) [40, 41, 44-46], or used a wide age range (1-14 years) [47, 48]. The studies that included young children (0-6 years) used observational methods in which behaviors were counted by frequency, time intervals, or the presence of a specific behavior [38, 49-53]. However, these types of observation methods have some disadvantages: they are time consuming and behaviors cannot be evaluated in the context of other behaviors [54-56]. Therefore, there is a need for an observational instrument, which is not time consuming and could evaluate observed behaviors in the context of other behaviors. With this type of observational instrument, researchers could examine whether certain parent and/or child behaviors are associated with child outcomes, like the HbA_{1c} level and QoL of the child. Therefore, we clearly described our plans a priori, on how to develop such an observation instrument (see **Chapter 3**) before we actually developed the observation instrument itself (see **Chapter 4**).

The association between parent-child interaction and child outcomes (HbA_{1c} level and quality of life)

There are a few studies that have focused on the quality of parent-child interactions in children with T1DM. The studies that have been conducted showed that over-involvement, restrictiveness, and hostility of the parent, conflicts and negative communication were associated with suboptimal HbA_{1c} levels [57-63] and a lower QoL of the child [45, 64]. On the other hand, positive communication, reinforcement, warmth, caring behavior, and emotional support were associated with more optimal HbA_{1c} levels [61, 62, 65] and a better QoL of the child [61, 66]. An important disadvantage of the aforementioned studies is that they did not include or included only a few (very) young children with T1DM. This means that research on the quality of the parent-child interaction in younger children and its potential associations with the HbA_{1c} level and QoL is lacking. Therefore, we have examined whether the quality of parent-child interaction was associated with the HbA_{1c} level and QoL in young children with T1DM in **Chapter 5**.

Parenting young children with type 1 diabetes and being responsible for their treatment is stressful

For children with T1DM (below the age of about 8 [39]) is it difficult to adequately deal with the disease themselves; therefore their parents bear the complete responsibility for the diabetes-management [48, 67]. Because of this full responsibility (assessing blood glucose levels, administering insulin, regulating

food intake, and guarding these parameters in conjunction with the level of physical activity of their child), having to think constantly about the correct amount of insulin/carbohydrates, and making adjustments during the day without any help of others, parents could experience this as having an 'extra job'. Therefore, it is not surprising that many parents of young children with T1DM report to experience high levels of parenting stress [68].

Parents of young children with T1DM report higher levels of parenting stress than parents of older children with T1DM [69-71]. This can be explained by the behavior of young children, as they can be unpredictable in their physical activity level and eating behavior and because they do not always express symptoms when having a high or low blood sugar level [72]. Parents of young children with T1DM, therefore, should always be vigilant [73], even during the night as children could experience hypoglycemic events [74, 75]. Another situation that is often regarded as stressful by parents is the mealtime [50]. At that time both food intake and insulin administration has to be managed, in addition to normal mealtime behaviors. [76]. Therefore, parents of children with T1DM do not only experience general parenting stress like many parents, but also disease-related parenting stress. Disease-related parenting stress, i.e. pediatric parenting stress, distinguishes from general parenting stress by taking into account specific disease-related factors like the child's health, treatment demands, communication with the child and medical team, emotional and role functioning [77].

Parenting stress is related with both medical and psychological outcome factors

Based on prior research, it can be hypothesized that a higher level of pediatric parenting stress could be related with the HbA_{1c} level of the child in a bi-directional way. High HbA_{1c} levels can cause parental stress, but on the other hand, a certain level of parental stress is perhaps needed to achieve and maintain a lower , more optimal, HbA_{1c}. Indeed, a study including parents of young children with T1DM has shown that a higher level of pediatric parenting stress was related with a more optimal HbA_{1c} level [71]. But again on the other hand, some parents feel distressed when their child has a relatively high blood glucose level as they strongly aim to avoid long- and short-term complications. Therefore, higher levels of pediatric parenting stress could also contribute to higher levels of parental involvement of diabetes-management and lead to more optimal HbA_{1c} levels in these young children [78]. This suggests that a certain amount of pediatric parenting stress seems to benefit the medical outcomes in young children with T1DM. However, there is only one study that has examined this association in young children with T1DM; in that particular study, all children received insulin

through multiple daily insulin injections [71]. Nowadays, however, most young children are on insulin pump therapy because of the benefits of using a pump [79]. As parents of young children with T1DM on insulin pump therapy report lower levels of (pediatric) parenting stress [69, 70], the association between parenting stress and HbA_{1c} levels in these children might be different. However, the association between parenting stress and HbA_{1c} levels in children with insulin pumps has not been examined yet.

Literature on the associations between parenting stress and QoL in young children with T1DM is also lacking. A study with older children (12-17 years) [80] found that higher levels of general parenting stress were associated with a lower reported child QoL. Maybe higher levels of general and pediatric parenting stress are also related to a lower child QoL in young children with T1DM as the stress affects the parent's and therefore the child's mood. Literature in other pediatric populations supports this assumption, for example, a study with children with cerebral palsy showed that higher levels of parental stress were associated with lower child QoL [81].

In **Chapter 5**, as mentioned earlier, we examine the associations between the quality of parent-child interactions and child outcomes (HbA_{1c} level and QoL). Because research has shown that a lower quality of parent-child interaction is strongly correlated with a higher level of parenting stress [for example 82, 83], we also believe that a higher level of parenting stress is related to child outcomes like a higher HbA_{1c} level and lower QoL of the child (or vice versa). Because of a lack in research on the associations between parenting stress and child outcomes (HbA_{1c} level and QoL), we also have examined these associations in **Chapter 5**.

Pediatric parenting stress in both fathers and mothers of young children with type 1 diabetes

In the field of pediatric parenting distress, fathers of young children with T1DM are understudied. For example, a recently published review [78] included only one study with fathers of young children (2-6 years) with T1DM. This study showed that fathers experienced relatively mild levels of pediatric parenting stress [72]. Unfortunately, no information about the pediatric parenting stress of the mothers was reported. Although it can be expected that mothers experience higher levels of pediatric parenting stress than fathers based on research in parents of older children with T1DM (7-14 years) [84, 85]. Furthermore, mothers are most of the times responsible for the diabetes-management of their child [86] and, therefore, might experience higher levels of pediatric parenting stress than their spouses. There seems to be a lack of information about the differences in pediatric parenting stress in mothers and fathers of young children with T1DM, while this evidence is needed to develop appropriate interventions and to be able

to provide adequate support for families.

Even though literature has shown that parents of young children with T1DM report higher levels of pediatric parenting stress [69-71], literature on pediatric parenting stress in these families over time is limited. This information, however, is needed to be able to provide the help and support these parents might need. As far as we know, only one study has examined pediatric parenting stress in parents of young children (4-7 years) with T1DM over time [69]. This study, however, included a small group of parents (n=24), and examined the change in pediatric parenting stress after their child has changed treatment regimen. Therefore, knowledge about the changes over time in pediatric parenting stress in parents of young children with T1DM without changing treatment regimen is lacking, but needed to be able to provide the support and help these parents might need. Therefore, we have compared the level of pediatric parenting stress between fathers and mothers of young children with T1DM and studied the course of pediatric parenting stress over time (see **Chapter 6**).

Attachment might act as a moderator on the association between pediatric parenting stress and child outcomes (child behavior problems, HbA_{1c} level, and diabetes-specific quality of life)

Research in children with T1DM has shown that parenting stress and child behavior problems are positively associated [87-89]. However, research in the general population has also shown that the quality of the attachment relationship acts as a moderator on this relationship between parenting stress and child behavior problems [90]. Attachment can be described as the emotional bond between an infant and his or her parent [91]. This bond develops over time [92], which is based on the daily parent-child interactions [93, 94]; higher quality of parent-child interactions represent a better attachment than lower quality of parent-child interactions [95]. In infancy, children become attached to their parents [96]. Infants form an attachment relationship with their parents, who function as a secure base for the child to explore the environment [97]. This attachment relationship between parent and child can be classified as 'secure' or 'insecure' [98]. Children who are securely attached explore the environment when the attachment figure is present, are upset for only a short period of time when the attachment figure leaves and show affection toward the attachment figure when he/she returns, while children who are insecurely attached do not explore the environment that much, are very upset or not upset at all when the attachment figure leaves and show resistance or avoidance toward the attachment figure when he/she returns [91, 99-101]. The nature of this attachment relationship has a strong influence on later child development [100,

102] and is therefore an important focus of interventions [91, 103].

Currently, attachment in young children with T1DM has not been examined. Previous research with healthy children has shown that the quality of the attachment relationship acts as a moderator on the relationship between parenting stress and child behavior problems [90], but this has not been studied in young children with T1DM. More specifically, we therefore aimed to examine whether the quality of attachment acts as a moderator on the relationship between pediatric parenting stress and child outcomes (child behavior problems, HbA_{1c} level, and diabetes-specific QoL) in young children with T1DM in **Chapter 7**.

Initiation of the OKI-DO study

The data for this thesis were collected within the OKI-DO study (Ouder Kind Interactie – Diabetes Onderzoek). All families with a young child (0-7 years) with T1DM from 15 hospitals in the Netherlands were invited to participate. Excluded were families with a child with a mental disability, Down syndrome, an Autism Spectrum Disorder, or when families lacked basic proficiency in Dutch. In total, 17 families were excluded. Of the remaining 121 families, 77 families (64%) agreed to participate in the OKI-DO study. All these families were contacted to make an appointment for a home visit. The home visit consisted of videotaping parent-child interactions and collecting the completed questionnaires (assessing (pediatric) parenting stress, (diabetes-specific) QoL, and child behavior problems) by both parents. Furthermore, after each home visit, the quality of the attachment relationship between caregiver and child was assessed and the HbA_{1c} level was retrieved from the medical records of the children. One year after the home visit, both parents were asked to complete the questionnaire about pediatric parenting stress again.

Summary of the aims of the study:

Figure 2 represents the present thesis in a structured way:

- The striped line represents the strong correlation between the quality of parent-child interaction and parenting stress, which already has been examined thoroughly for example [82, 83]. The grey line represents the attachment theory, which states that the quality of the attachment relationship is positively associated with the quality of parent-child interaction [93, 94]. The striped and grey line have been outlined in the present chapter (**Chapter 1**).
- In **Chapter 2**, we focus on one specific child outcome, namely the QoL of children with T1DM (box *child outcomes* in Figure 2). The aim of this chapter is to gain more insight into the quality of life of children with T1DM.
- In **Chapter 3 and 4** the development of an observation rating scale to assess

parent-child interaction during diabetes-specific situations is described (box *parent-child interaction*).

- **Chapter 5** is represented in the dotted lines. The aim is to examine relationships between the quality of parent-child interaction and parenting stress with the HbA_{1c} level and Qol of the child.
- The box *parenting stress* is further examined in **Chapter 6**, in which we will gain more insight into pediatric parenting stress of both fathers and mothers of young children with T1DM.
- The role of attachment is represented in the black line. In **Chapter 7** we will examine whether the attachment relationship acts as a moderator on the relationship between pediatric parenting stress and child outcomes (child behavior problems, QoL, and HbA_{1c} level).

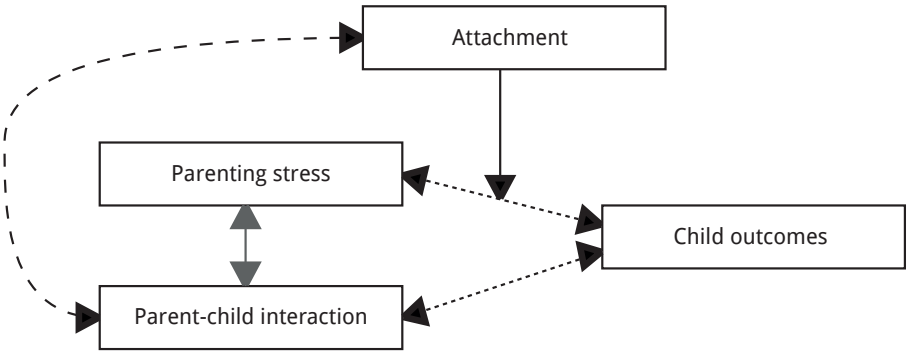

Figure 2: Overview of the present thesis.

An overview of the main findings of the present thesis will be discussed in the last chapter, in which we will also critically review the OKI-DO study, suggest practical and clinical implications, and recommend possible future directions (see **Chapter 8**).

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Chapter 2

Quality of life of children with type 1 diabetes: a systematic review

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ABSTRACT

Introduction
Children with type 1 diabetes mellitus (T1DM) have to deal with a complex and demanding daily treatment regime which can have a negative impact on the quality of life (QoL) of these patients. The objective of the present study is to review studies that have compared generic quality of life of children and adolescents with T1DM with that of healthy peers. In addition, we will examine whether QoL differs between boys and girls, and across different developmental stages.

Methods
A systematic literature search using PubMed was conducted for the years 2000 through May 2012. 17 studies were eligible for the current review. Effect sizes were computed to estimate the effects of having T1DM on QoL in children and adolescents.

Results
Although individual studies reported small to moderate effect sizes on the distinct QoL-domains, the weighted effect sizes across all studies indicated no differences in QoL-domains between children and adolescents with T1DM and healthy controls. However, disease-specific problems were certainly present. Girls with T1DM reported lower generic and disease-specific QoL than boys with T1DM. Relationships between age and generic or disease-specific QoL remained unclear.

Conclusions
Although children and adolescents with T1DM have to live with a demanding treatment regime, overall results revealed that their generic QoL is not impaired compared to healthy peers. However, disease-specific QoL problems, including a negative impact of diabetes on daily functioning, and diabetes-related worries were certainly present. Longitudinal research is needed in order to provide tailored care for children of all ages with T1DM.

Introduction

Results of the Diabetes Control and Complications Trial (DCCT) have convincingly shown that keeping blood glucose levels close to normal levels avoids or delays the onset of long-term complications of diabetes [1]. To achieve or maintain optimal glycemic control, children with type 1 diabetes (T1DM) and their parents have to deal with a complex and demanding daily treatment regime, including blood glucose monitoring (several times a day) and administering a correct dose of insulin at the right time, regulation of food-intake, and guarding these parameters in conjunction with their level of physical activity. Hypoglycemic events, the intensive and demanding treatment regime and changing blood glucose levels could impair the quality of life (QoL) of children and adolescents with T1DM [2, 3]. However, it has also been noted that children with T1DM do not necessarily report lower QoL and sometimes even show adaptive outcomes [4, 5].

Obviously, the first goal of healthcare providers is to optimize metabolic control of children with T1DM. However, nowadays, healthcare providers are more and more challenged to guard and maintain the QoL of these children because QoL has become clearly established as an important endpoint in medical care [6]. This especially applies to chronic diseases for which complete recovery is unlikely. Although definitions of QoL vary widely, there is consensus about two central aspects. First, QoL should be regarded as a multidimensional construct incorporating at least three broad domains that can be affected by one's disease or treatment, including physical, mental and social functioning [7, 8]. Second, QoL should be assessed from the patient's perspective whenever possible [8-11]. Recommendations with respect to the minimum age of children to complete QoL instruments vary from seven to nine years [11], parents often serve as proxies for younger children [12].

Most instruments to assess QoL can be classified as either generic or disease-specific. The generic instruments are designed to measure all aspects of health and well-being irrespective of the underlying disease, and enable comparisons across different disease groups, and with healthy or non-diabetic comparison groups. However, the generic instruments are limited in that they do not measure specific aspects that are of particular relevance to patients with T1DM, such as disease symptoms or the impact of certain treatments. Conversely, disease-specific measures examine the symptoms of specific disease groups and how they function [13, 14].

Several narrative reviews have studied (aspects of) the QoL of children with T1DM [15-19]. The results showed that children and adolescents with T1DM are more likely to experience psychosocial problems such as depression, behavioral problems and anxiety than healthy peers [15-19]. However, none of these reviews has been conducted systematically. To our knowledge, there is only

one systematically conducted review [20] which, however, focused only on mental QoL, leaving out social, physical, and disease-specific QoL. This meta-analytic review [20] reports that children with T1DM are slightly at more risk for anxiety and depression, compared with comparison groups, although this effect was smaller among more recent studies [20]. A systematic overview of generic QoL in combination with disease-specific QoL in children and adolescents with T1DM worldwide is lacking, indicating that the complete impact of T1DM on QoL is unknown.

By reviewing the available literature, we attempt to maximize the information concerning generic QoL-domains (i.e., mental, social, and physical functioning) and disease-specific QoL (i.e., symptoms, worries, and disease-related problems) of children with T1DM.

Because girls are known to report their QoL lower as boys in the general population and in other patients groups [21, 22], and because children and adolescents have different developmental tasks (making friends in childhood versus being more independent in adolescence [23]) and therefore might perceive their QoL differently, we also decided to examine the potential effects of gender and age on QoL in the included studies. Therefore, our review has four objectives:

1. to compare generic QoL-domains of children and adolescents with T1DM with healthy controls,
2. to examine whether QoL differs between boys and girls with T1DM,
3. to examine whether QoL of patients with T1DM differs across different developmental stages (e.g., children versus adolescents),
4. to examine which diabetes-specific QoL domains are most affected in children and adolescents with T1DM.

Methods

Search strategy

A literature search was conducted using the PubMed database for the years 2000 through May 2012. We decided to exclude articles published before 2000, because of substantial changes in treatment regimen and new clinical standards in the last few years [24, 25].

A combination of the following entries in title and/or abstract was used: 'type 1 diabetes' or 'type 1 diabetes mellitus' or 'juvenile-onset diabetes' or 'juvenile-onset diabetes mellitus' or 'insulin dependent diabetes' or 'insulin dependent diabetes mellitus' or 'diabetes type 1'or 'diabetes mellitus type 1' in combination with 'quality of life', or 'health status' or 'social' or 'psychological' or 'psychosocial' or 'mental', and 'child' or 'children' or 'youth' or 'adolescents' or 'adolescent', and 'healthy control', or 'control', or 'control group', or 'comparison', or 'comparison group', or 'general', or 'general population', or 'population'. It

was also stated that keywords in title/abstract should not include, 'pregnancy', 'genetics', 'transplantation', 'chromosome', 'islet functioning', 'medicine', 'type 2'. While searching, two limits were activated: results should only include articles published in the year 2000 or later, and written in the English language.

Statistics

First, to compare generic QoL of groups of patients with T1DM with healthy controls, we calculated effect sizes, Hedges' d, based on standardized differences between mean scores, t-statistics or correlations [26]. Separate weighted effect sizes *d+* across all included studies, which weigh the effect sizes according to the respective sample sizes [26, 27], were calculated for physical QoL, psychosocial QoL, and overall QoL. According to Cohen, effect sizes *d* of 0.20, 0.50, and 0.80 can be considered small, medium, and large, respectively [28].

When the included studies compared generic and/or diabetes-specific QoL between boys and girls with T1DM and across different age groups with T1DM, effect sizes were also calculated. When the available studies did not include data to calculate effect sizes, results of gender and age differences were described.

To be able to compare (outcomes of) the studies, each study was evaluated with an evaluation score. The following criteria were developed to evaluate the quality of the study. Criterion A: assessing all three (2 points), two (1 point) or one (0 points) QoL-domain(s). Criterion B: assessing both generic and disease-specific (1 point), or only one type of QoL (0 points). Criterion C: assessing QoL with self-reports (2 points), partly self-reports (1 points), or only proxy reports (0 points), except when patients were too young for self-reports [11, 12]. Criterion D: sample with more than 100 (3 points), between 51 and 99 (2 points), between 10 and 50 (1 point), or less than 10 patients (0 points). Criterion E: results between different age groups with T1DM were presented separately (1 point), or not (0 points). Criterion F: results between boys and girls with T1DM were presented separately (1 points), or not (0 points). Hence, scores for evaluating the included studies could vary from 0 to 10.

The first search resulted in 298 hits. Studies were included when they (a) were empiric quantitative studies, (b) focused on at least one of the domains of generic QoL and used a QoL-questionnaire to assess QoL, (c) included a healthy control group, and (d) provided statistics to calculate effect sizes. The flow chart in Figure 1 shows the search and selection of relevant studies. Based on the inclusion criteria, 18 studies were included [3-5, 29-43]. An additional manual search of the references of the selected articles and previous narrative reviews was carried out, which resulted in the inclusion of no more articles. Two studies [29, 40] did not report mean scores, t-statistics or correlations, effect sizes of

these studies were derived from figures in the articles. One study by Varni et al. [43] was excluded because it was based on data from another study, that was already included. Therefore, a total of 17 studies were included in this review [3-5, 29-42].

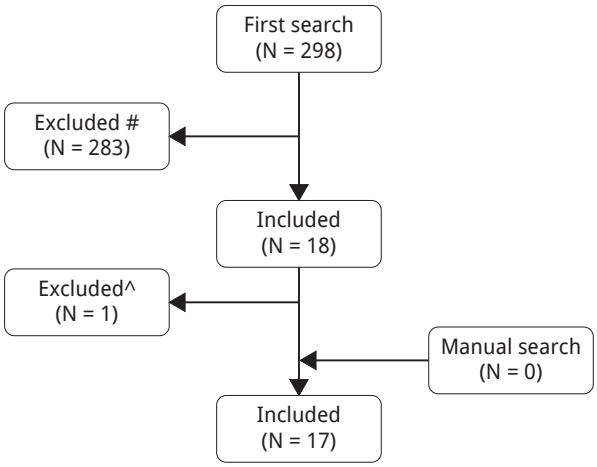

Figure 1: Flow chart of literature search and selection.

 # Studies that had no healthy comparison groups when discussing generic QoL or which mainly examined the following topics were excluded: reviews, QoL of parents or family, family functioning, intelligence, qualitative studies, other diseases or T1DM in combination with T2DM or other diseases. When articles tested the effectiveness of interventions or treatments and also provided QoL-measures at baseline, they were included.
^ One study by Varni et al. [43] was excluded because of the use of the same diabetes sample in another included article [38]; we decided to use the most recent one [38].

Results

Characteristics of included studies

An overview of the included studies, their characteristics, and evaluation is presented in Table 1. Studies differed considerably on several features. Sample sizes ranged from N=68 to N=629, the percentage of male participants ranged from 38% to 57% (when data was available); mean age in the studies ranged from 10 to 15 years (range 2-19 years). When studies included both self- and proxy-reports in assessing child's QoL and calculating effect sizes was possible for both assessments, only the children's perspectives were taken into account.

Of the 17 included studies, eight studies used the PedsQL for assessing generic QoL [3, 4, 30, 33, 34, 37-39]. Other instruments used to assess QoL were the CHQ-CF [32, 35, 40], the KINDL-R [29, 41], the MMQL [42], the DQoL Measure for Youth [31], a Japanese HRQoL questionnaire [5], or a specially designed single-item question [36].

Of the 17 included studies, six studies assessed disease-specific QoL with all of them using validated QoL-questionnaires including the PedsQL Diabetes Module [3, 4, 30], the Diabetes Quality of Life questionnaire [32], the Diabetes Related-Quality of Life [5], or the DQoL Measure for Youth [31].

The included studies were also evaluated at 6 criteria (see *statistics*). Scores for evaluating the included studies could vary from 0 to 10. Table 1 shows the evaluation scores of all included studies.

Comparison between children with T1DM and healthy controls in generic QoL domains

Effect sizes (Hedges' d) of physical, psychosocial, and overall QoL are shown in Table 1. Effect sizes were first calculated per subscale of the used instruments in each study. After that, mean effect sizes of physical, psychosocial, and overall QoL were calculated for each study by including the effect sizes of scales that assessed this QoL-domain. For example, in the study of de Wit et al. [40] the effect size of physical QoL (*d = -0.12*) consisted of the effect sizes of the subscales 'physical functioning' (*d = 0.00*), 'role-functioning physical' (*d = -0.31*) and 'bodily pain' (*d = -0.05*). The effect size of psychosocial QoL (*d = -0.05*) was calculated by taking into account the effect sizes of the subscales 'role-functioning emotional' (*d = -0.11*), 'role-functioning behavioral' (*d = 0.07*), 'behavior' (*d = 0.00*), 'mental health' (*d = -0.07*), 'self-esteem' (*d = 0.00*), and 'family cohesion' (*d = -0.06*). The effect size of overall QoL (*d = -0.36*) consisted of the effect size of 'general health' (*d = -0.36*).

The weighted effect sizes[1] (not given in Table 1) were -0.05 (n=14) for physical QoL, -0.00 (n=14) for psychosocial QoL and -0.05 (n=15) for overall QoL.

Hence, although individual studies reported small to moderate effect sizes on the distinct QoL-domains, the weighted effect sizes d+ across all studies indicated minimal differences in QoL-domains between children and adolescents with T1DM and healthy controls.

Comparison between boys and girls with T1DM in QoL

Of the 17 included studies, there were only two studies [3, 32] that compared generic QoL and three studies [3, 31, 32] that compared disease-specific QoL between boys and girls with T1DM that allowed calculating effect sizes of the gender differences. Positive effect sizes indicate that boys reported better generic QoL than girls (not shown in table). Effect sizes based on the results of these studies were 0.19 for physical QoL [32], 0.29 [32] for psychosocial QoL, and 0.28 [32], and 0.51 (Cohen's d) [3] for total QoL. These effect sizes, with small or medium effects, indicate that girls reported lower generic QoL as compared to boys.

[1] *Only Hedges'd effect sizes were taken into account when calculating the weighted effect size.*

For disease-specific QoL, the effect sizes were 0.36 for diabetes-related worries [32], 0.31 for diabetes impact [32], 0.44 [32], 0.52 (Cohen's d) [31] for satisfaction, and 0.47 (Cohen's d) [3] for overall disease-specific QoL. These effect sizes indicate that girls reported having more worries about diabetes, less satisfaction and lower overall diabetes-specific QoL than boys. Unfortunately, the small number of studies prohibited the calculation of weighted effect sizes.

When the calculation of an effect size was not possible, for example due to the lack of means and standard deviations, results of individual studies showed similar results [4, 29, 40], except for one study who found no significant differences between boys and girls [36].

Comparison between different age groups of patients with T1DM

Of the 17 included studies, there were only four[2] studies that compared generic QoL [3, 29, 32, 35] and only one study that compared diabetes-specific QoL [3] between different age groups with T1DM that allowed calculating effect sizes. Positive effect sizes indicate that older children reported better generic QoL than younger children (not shown in table). Effect sizes based on the results of these studies were 0.12 [32], -0.03 [35], and -0.09 [29] for physical QoL, -0.36 [32], -0.09 [35], and -0.26 [29] for psychosocial QoL, and -0.25 [32], -0.22 (Cohen's d) [3], -0.01 [35], and -0.46 [29] for total QoL. These effect sizes, ranging from small to medium, indicate that older children (aged 12-18 [35], 13-16 [29], 13-18 [3], and 15-18 [32]) reported lower generic QoL as compared to younger children (aged 5-11 [35], 8-12 [3, 29], and 11-14 [32]).

For diabetes-specific QoL, the effect size was calculated at 0.47 (Cohen's d) for overall diabetes-specific QoL [3], indicating that children (aged 8-12) reported lower disease-specific QoL and more problems than adolescents (aged 13-18). Unfortunately, the small number of studies prohibited the calculation of weighted effect sizes.

When the calculation of an effect size was not possible, due to the lack of the needed statistics, results of individual studies showed no clear results. Some studies found no differences in generic and/or diabetes-specific QoL in different age groups [4, 34, 40], while others did found an effect of age on generic and/or diabetes-specific QoL [4, 31, 32, 36]. One study used a different questionnaire in the younger (primary/junior school) and older (high school) group children, so comparing these different age groups was unfortunately not possible [5].

2 The study of Wake [35] only reported data on the CHQ-Parent From on which we could calculate effect sizes between different age groups.

Diabetes-specific Quality of Life

Of the 17 included studies, six studies examined diabetes-specific QoL [3-5, 30-32]. The two most affected domains in the PedsQL are: Worry and Treatment barriers [30], Diabetes symptoms and Worries [4], Communication and Treatment barriers [3]. The two most affected domains in other studies are: Family burden related to diabetes and Family involvement [5], Impact and Satisfaction [32], Disease Impact and Disease-Related Worries [31].

Table 1: Study characteristics and effect sizes for comparisons between patients with type 1 diabetes and healthy controls.

Study	a. TIDM sample b. Healthy sample	Instruments	Relevant outcome variables	Effect sizes				Evaluation score
				d	Physical QoL	Psychosocial QoL	Overall QoL	
1. De Wit et al. [40]	a. n=91, 13-17 years, 52% male b. n=466, 9-17 years, 51% male	CHQ-CF (87 items) data on the subscale Family activities was not available	Physical functioning Role-functioning emotional Role-functioning behavioral Role-functioning physical Bodily pain Behavior Mental health Self-esteem General health Family cohesion	0.00 -0.11 0.07 -0.31 -0.05 0.00 -0.07 0.00 -0.36 -0.06	-0.12	-0.05	-0.10	8
2. Emmanouilidou et al. [4]	a. n=84, 2-18 years, 53% male b. n=89, 2-18 years, 53% male	PedsQL	Physical health Psychosocial health Total score	0.07 0.16 0.06	0.07	0.16	0.06	9
3. Faulkner [31]	a. n=69, 13-19 years, 54% male b. n=75, 13-19 years, 35% male	DQoL	Life satisfaction subscale	-0.40#	-	-	-0.40#	8
4. Graue et al. [32]	a. n=63, 15-18 years, 52% male b. n=38, 15-18 years, 58% male	CHQ-CF (87 items) only data of children aged 15-18 could be compared	Physical functioning Role-functioning emotional Role-functioning behavioral Role-functioning physical Bodily pain Behavior Mental health Self-esteem General health Family activities Family cohesion	0.13 -0.07 -0.10 0.03 0.35 -0.28 -0.08 -0.14 -0.48 -0.04 -0.07	0.17	-0.11	-0.07	10

Table 1 continued

5. Hutchings et al. [42]	a. n=73, 8-18 years, NA b. n=563, 8-18 years, NA	MMQL	Appearance School functioning Social functioning Emotional functioning Physical functioning	0.20 0.12 0.18 0.07 0.05	0.13	0.12	0.13	6
6. Jafari et al.[30]	a. n=94, 8-18 years, 44% male b. n=200, 8-18 years, 47% male	PedsQL	Physical health Psychosocial health Total score	-0.50 -0.49 -0.50	-0.50	-0.49	-0.50	7
7. Kalyva et al. [3]	a. n=117, 5-18 years, 45% male b. n=128, 5-18 years, 51% male	PedsQL	Physical HRQoL Psychosocial HRQoL Total generic HRQoL	-0.43# -0.24# -0.34#	-0.43#	-0.24#	-0.34#	10
8. Laffel et al. [34]	a. n=100, 8-17 years, 53% male b. n=401, normative sample	PedsQL	Physical subscale Psychosocial subscale Total score	0.09 -0.06 0.00	0.09	-0.06	0.00	8
9. Maas-van Schaaijk et al. [36]	a. n=151, 12-18 years, 43% male b. n=122, 12-18 years, 41% male	Single item questionnaire	Total QoL	-0.31	-	-	-0.31	7
10. Nakamura et al. [5]	a. n=368, 9-18 years, 38% male b. n=368, 9-18 years, 38% male	Japanese questionnaire for primary/junior school children Japanese questionnaire for high school children	Anxiety Family Friends School General health Strength/diligence/self-esteem Total score Friends School Mental health Parent/economy state Relationship with friend/self-esteem Vitality Anxiety (school/employment) Siblings Total score	0.19 0.08 0.09 0.20 -0.02 0.24 0.20 0.04 0.23 0.02 -0.11 0.03 0.03 0.17 -0.09 0.08	0.03	0.12	0.10	9

Table 1 continued

11. Nardi et al. [37]	a. n=70, 6-18 years,56% male b. n=70, 6-18 years, 57% male	PedsQL	Physical functioning Psychosocial functioning (only data on the emotional subscale were provided) Total QoL	-0.06 -0.18 -0.26	-0.06	-0.18	-0.26	6
12. Stahl et al. [41]	a. n=629, 11-17 years, 54% male b. n=6813, 11-17 years, 51% male	PKINDL-R	Physical well-being Emotional well-being Self-esteem Family Friends School Total score	-0.05 -0.06 0.20 -0.15 0.01 0.15 0.03	-0.05	0.11	0.03	7
13. Upton et al. [33]	a. n=124, 8-18 years, NA b. n=1033, 8-18 years, NA	PedsQL	Physical subscale Psychosocial subscale Total score	-0.07 0.04 0.01	-0.07	0.04	0.01	7
14. Varni et al. [38]	a. n=209, 5-19 years, 48% male b. n=5480, 5-19 years, 52% male	PedsQL	Physical subscale Psychosocial subscale Total score	-0.13 -0.05 -0.15	-0.13	-0.05	-0.15	7
15. Varni et al. [39]	a. n=92, 5-18 years, 53% male b. n=695, 5-18 years, 51% male	PedsQL only data on the paper administration could be compared	Physical subscale Psychosocial subscale Total score	-0.37 -0.43 -0.44	-0.37	-0.43	-0.44	6
16. Wagner et al. [29]	a. n=68, 8-16 years, 57% male b. n=1502, 8-16 years, 52% male	KINDL-R	Physical well-being Psychological well-being Self-esteem Family Friends School Total QoL	0.15 0.14 0.11 0.12 -0.03 0.35 0.17	0.15	0.15	0.17	8

Table 1 continued

17. Wake et al. [35]	a. n=128, 5-18 years, 56% male b. n=5414, 5-18 years, 50% male	CHQ-CF (80 items) only data of children aged 12-18 could be compared	Physical functioning Role-functioning emotional Role-functioning behavioral Role-functioning physical Bodily pain Behavior Mental health Self-esteem General health Family activities Family cohesion	0.03 0.01 -0.02 -0.09 0.08 0.00 0.09 0.01 -0.29 -0.19 -0.09	0.07	-0.06	-0.07	7

 # For two studies effect size Cohen's d was calculated based on t-values and F-values, because they did not provide means and standard deviations to calculate Hedges'd These effect sizes were not included in the calculation of the weighted effect sizes
Effect sizes were (re)coded so that negative effects indicated that disease groups did worse than the healthy groups and positive effects indicated that disease groups did better than healthy groups
The following criteria were developed to evaluate the quality of the study. Criterion A: assessing all three (2 points), two (1 point) or one (0 points) QoL-domain(s). Criterion B: assessing both generic and disease-specific (1 point), or only one type of QoL (0 points). Criterion C: assessing QoL with self-reports (2 points), partly self-reports (1 points), or only proxy reports (0 points), except when patients were too young for self-reports [11, 12]. Criterion D: sample with more than 100 (3 points), between 51 and 99 (2 points), between 10 and 50 (1 point), or less than 10 patients (0 points). Criterion E: results between different age groups with T1DM were presented separately (1 point), or not (0 points). Criterion F: results between boys and girls with T1DM were presented separately (1 points), or not (0 points). Scores for evaluating the included studies could vary from 0 to 10.
Abbreviations: CHQ-CF = Child Health Questionnaire – Child Form; DQoL = Diabetes Quality of Life Measure for Youth; KINDL-R = KINDer Lebensqualitätsfragebogen Revised; MMQL = Manchester-Minneapolis Quality of Life instrument; PedsQL= Pediatric Quality of Life Inventory

Discussion

The first objective of this systematic review was to compare generic QoL of children and adolescents with T1DM with that of healthy comparison groups. On average, children and adolescents with T1DM showed no differences in physical, psychosocial, and overall QoL as compared to healthy peers. This finding is not in line with results of previous, narrative reviews, which reported that children and adolescents with T1DM reported more psychosocial problems and lower QoL than healthy peers [15-19]. However, these narrative reviews were conducted non-systematically, focused mainly on just one part of QoL, like psychological problems [16], psychosocial problems [17, 19], or depression [18], and were conducted (almost) more than 10 years ago [15, 18, 19]. It seems plausible that the different conclusions may be a result of substantial improvements in treatment regimen, as the recent meta-analysis about depression among children with T1DM found that group differences are getting smaller with time, which is consistent with our findings [20].

In the current systematically conducted review, some individual studies showed lower reported QoL for children and adolescents with T1DM. When the weighted effect sizes d+, that take sample sizes into account, across all included studies, were calculated, it appeared that on average studies did not show any QoL differences between patients with T1DM and healthy peers. However, it should be mentioned that the included studies used different instruments to measure QoL, which could influence the results. To be able to determine which outcomes of individual studies were most valuable, we evaluated the methodological quality of each included study (see Table 1). The four high quality studies with an evaluation score of 9 or 10 [3-5, 32], did not show consensus, as two studies [3, 32] showed a negative effect of T1DM on QoL and the two other studies [4, 5] did not. As the way of determining QoL, included QoL domains, and sample size, were included in the composition of the evaluation scores, a possible explanation of these contrasting findings remains unclear.

Although we found that, on average, no differences were found in generic QoL domains between children with T1DM and healthy controls, several studies reported that disease-specific problems were certainly present [3-5, 30, 32]. For example, children and adolescents with T1DM worried about possible consequences of their disease (e.g., worries about whether they would pass out), were less satisfied with their social and school life (e.g., less satisfaction with social relationships and friendships), felt like their disease was a burden on their families, and reported that T1DM influenced their daily functioning (e.g., diabetes interrupted their leisure time activities) [3-5, 30, 32].

Furthermore, it was found that girls perceived lower generic QoL than boys [3, 29, 32, 40]. The studies examining gender differences in diabetes-specific

QoL found the same pattern, as girls consistently reported more diabetes-related worries and lower overall disease-specific QoL than boys. It should be noted, however, that studies with other patient groups, or healthy children, also showed that QoL was poorer for girls [21, 22]. Apparently, girls perceive more QoL problems than boys in general, independent of their health status. The Hvidore Study Group, who found the same result in 2101 adolescents with T1DM [44], also formulated some diabetes-related hypotheses about why girls report more QoL problems than boys with T1DM. This difference could be related to the earlier onset of puberty and hormonal changes in girls [45-48], and with abnormal eating behaviors and lack of physical activity in girls [49, 50] as explained by the Hvidore Study Group [44].

Results of the studies that examined to which extent generic and disease-specific QoL differed across developmental stages, revealed no clear patters. It was difficult to group the patients according to mutually exclusive age groups, because age ranges varied across the included studies, which hampered comparison of studies. For example, in the study of Wagner et al. [29] patients aged between 8 and 12 years were designated as children and patients aged between 13 and 16 years as adolescents, whereas in the study of Graue et al. [32] patients aged between 11 and 14 years were designated as younger adolescents and aged between 15 and 18 years as older adolescents. Furthermore, most studies examining QoL differences across developmental stages have been conducted with children and adolescents over 8 years, which precluded us to draw conclusions about the prevalence of generic and disease-specific problems in young patients. Therefore, the results of the current review might be more applicable to older children (>8 years) and adolescents having T1DM. Hence, specific knowledge of the QoL of the youngest patient-group is lacking, and no specific statements can be made about the QoL of a young child with T1DM. To summarize, most studies found that younger children with T1DM reported better generic QoL than older children with T1DM. One explanation of this finding might be due to higher levels of stress in the adolescent group [51]. Furthermore, one study [3] looked at diabetes-specific QoL and found that older children reported better disease-specific QoL compared to younger children. This could be explained by the fact that older children might be more habituated to their treatment and more independent of their parents in respect of their diabetes-management [52].

Limitations

A problem when reviewing the generic and disease-specific QoL of children and adolescents with T1DM is the heterogeneity of the study methodology between studies. Not all studies included in the current review did

employ the above consensus definition of QoL and used validated generic QoL questionnaires. The use of different questionnaires to assess generic QoL or one of its aspects might explain why different results were found between individual studies. Other limitations were that studies used selective samples (e.g. limited by age) or that statistics were often not calculated per age group but averaged across age groups. Another problem (as with all reviews) is that not all manuscripts are submitted for publication or accepted for publication and this may lead to publication bias. Selection bias may have occurred when published manuscripts were excluded because they did not report sufficient information to calculate effect sizes.

Future research

None of the included studies made use of a longitudinal study design: all of the included studies compared QoL between children in different developmental stages (i.e. made use of between-subjects design) rather than comparing QoL of the same children across different developmental stages (i.e. within-subjects design). In order to provide tailored care to help patients and their parents to cope with their disease, more knowledge about the course of QoL of children and adolescents with T1DM is needed to investigate the course of both generic and disease-specific QoL. Consequently, future research should also assess the QoL of (very) young children (< 8 years) with T1DM because these young children are often not included and information about the generic and diabetes-specific QoL in this group is scarce.

Furthermore, other variables, besides gender and age, should be taken into account, like glycemic control (HbA_{1c}), or time since diagnosis, as they might influence QoL [53, 54].

Clinical implications

Pediatricians who treat children and adolescents with T1DM should be well aware that although, on average, no generic QoL problems are prevalent (keeping in mind individual variation), disease-specific problems certainly are. They should also be aware that girls are at more risk for QoL problems than boys. Clinicians should not only inquire about patients' general wellbeing, but also disease-specific wellbeing. Because the results of the current review showed that the prevalence of disease-specific problems is very diverse, healthcare providers best provide treatments specifically tailored to individual patients. When clinicians are aware of additional diabetes-specific problems, they could refer their patients for further support and treatment, such as to other medical healthcare practitioners (e.g. dieticians, district nurses), to non-medical professionals who provide psychosocial support (e.g. psychologists, social

workers), or to a combination of health care providers. Furthermore, healthcare providers should periodically assess and discuss the QoL of their patients which has proved beneficial for the QoL of adolescents with T1DM [55].

Conclusion

Although children and adolescents with T1DM have to live with a demanding treatment regime and are at risk for complications and fluctuating blood-glucose levels, results of this review revealed that, on average, their generic QoL is not impaired as compared to healthy peers. However, studies reported that disease-specific QoL problems, such as negative impact of diabetes on daily functioning, diabetes-specific worries, and less satisfaction with life were prevalent in patients, although the prevalence of these problems varied between studies. Furthermore, girls with T1DM reported lower generic and disease-specific QoL than boys with T1DM, but this is also present in other patient groups and groups of healthy children. No clear association was found between age and generic and disease-specific QoL. Studies assessing generic and disease-specific QoL of very young children with T1DM as well as studies comparing generic QoL between different age groups are limited. More longitudinal research is needed in order to provide tailored care for children of all ages with T1DM.

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Chapter 3

Quality of the parent-child interaction in young children with type 1 diabetes: study protocol

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ABSTRACT

Introduction

In young children with type 1 diabetes mellitus (T1DM) parents have full responsibility for the diabetes-management of their child (e.g. blood glucose monitoring, and administering insulin). Behavioral tasks in childhood, such as developing autonomy, and oppositional behavior (e.g. refusing food) may interfere with the diabetes-management to achieve an optimal blood glucose control. Furthermore, higher blood glucose levels are related to more behavioral problems. So parents might need to negotiate with their child on the diabetes-management to avoid this direct negative effect. This interference, the negotiations, and the parent's responsibility for diabetes may negatively affect the quality of parent-child interaction. Nevertheless, there is little knowledge about the quality of interaction between parents and young children with T1DM, and the possible impact this may have on glycemic control and psychosocial functioning of the child. While widely used global parent-child interaction observational methods are available, there is a need for an observational tool specifically tailored to the interaction patterns of parents and children with T1DM. The main aim of this study is to construct a disease-specific observational method to assess diabetes-specific parent-child interaction. Additional aim is to explore whether the quality of parent-child interactions is associated with the glycemic control, and psychosocial functioning (resilience, behavioral problems, and quality of life).

Methods / Design

First, we will examine which situations are most suitable for observing diabetes-specific interactions. Then, these situations will be video-taped in a pilot study (N=15). Observed behaviors are described into rating scales, with each scale describing characteristics of parent-child interactional behaviors. Next, we apply the observational tool on a larger scale for further evaluation of the instrument (N=120). The parents are asked twice (with two years in between) to fill out questionnaires about psychosocial functioning of their child with T1DM. Furthermore, glycemic control (HbA_{1c}) will be obtained from their medical records.

Discussion

A disease-specific observational tool will enable the detailed assessment of the quality of diabetes-specific parent-child interactions. The availability of such a tool will facilitate future (intervention) studies that will yield more knowledge about impact of parent-child interactions on psychosocial functioning, and glycemic control of children with T1DM.

Introduction

Results of The Diabetes Control and Complications Trial (DCCT) have convincingly shown that keeping blood glucose levels close to normal levels avoids or delays the onset of long-term complications of diabetes [1]. When young children are diagnosed with type 1 diabetes mellitus (T1DM), parents get full responsibility for the diabetes-management of their child (e.g., blood glucose monitoring and administering insulin, regulation of food intake, and guarding the level of physical activity of their diabetic child). Normal and age appropriate behaviors that occur in the toddler and pre-school years (e.g. independence-seeking, refusing food, oppositional behavior) can interfere with the ability of parents to complete the tasks needed to achieve optimal blood glucose control [2]. This interference and the full responsibility of the parents may affect family functioning and parent-child interaction [3-5].

An overview by Anderson et al. [6] showed that when a child suffers from a chronic condition the parent-child relationship could be affected. Most studies described in this overview have shown negative effects of a medical condition on the quality of parent-child interactions, such as more conflict situations and less solution-directed communication, less cohesion, decreased medication adherence, and impaired functioning within the family [6]. For example, children with congenital heart disease reacted less responsive and their mothers appeared less sensitive than children and mothers in healthy families [7]. In families with a disturbed parent-child interaction, children with various somatic diseases showed more behavioral problems, but also more disease-related outcomes, such as an increased mean glycemic control (HbA_{1c}) in adolescents with diabetes [8]. Higher glycosylated hemoglobin is associated with more behavioral problems in youth with type 1 diabetes [9]. Because of these possible behavioral problems, parents want to keep their child's blood glucose values as close to normal as possible, to avoid the direct negative effect on the behavior of their child. To achieve this, the parents might need to negotiate with their child on the diabetes management tasks, but diabetes treatment is non-negotiable. These negotiations could negatively affect the interaction between parent and child. Diabetes is a 24/7 disease, so struggles on treatment tasks are not comparable with other (chronic) diseases.

Because parents are responsible for the treatment of their young child with T1DM and the child is fully dependent on his or her parents, we expect that the quality of parent-child interaction significantly contributes to both the psychosocial development and the quality of life of these children.

Given the importance of the topic, it is surprising that studies examining the quality of the parent-child interaction in families with young children with T1DM are scarce. Furthermore, the small number of studies that are available has

several shortcomings. For example, most studies examining the quality of parent-child interaction or related topics, have been conducted with older children with T1DM (> 8 years) [3, 4, 8, 10, 11], or used a wide age range (from 1 - 14 years [12, 13]). As a result, specific knowledge of the quality of the parent-child interaction of the youngest patient group is lacking, and no specific statements can be made about the quality of parent-child interaction in families with a young child with T1DM.

A second shortcoming is that in these studies self-report measures or semi-structured interviews were used [3, 4, 11-14]. In vivo observations and observational methods, however, appear more sensitive to subtle differences in family interactions [15]. These differences in interaction patterns may provide important implications for improving the quality of parent-child interactions during diabetes-management. Moreover, self-report measures and interviews reflect a subjective view from the perspective of parents, while by using observational methods, the interaction patterns can be assessed more objectively.

To our knowledge, there is only one research group [2, 16-19], that has studied the quality of parent-child interaction in young children with T1DM using an observational method. However, the studies of this research group only focused on a single dimension of a disease-specific parent-child situation, namely behavioral problems during the meal, while in fact the combination of diabetes-specific actions and behaviors around mealtime will give a more complete illustration of the diabetes-specific interactions (i.e. blood glucose monitoring, carbohydrate counting, and administering insulin).

Moreover, the observations were performed with an observational method [20] in which the behaviors of parents and children during the meal were to be counted (e.g., how often the child was encouraged to keep eating). This has an important disadvantage. In behavior counting methods, where all behaviors are counted, applying nuances is difficult, while with the so-called “rating scales” specific behaviors can be grouped under broad categories. This way of coding observational data provides room to make many dimensions and nuances in behaviors. Moreover, the predictive value of global rating scales has proved to be more appropriate than just counting specific behaviors [15, 21]. An additional advantage of rating scales is that it costs up to 5 times less time than counting all behaviors [15]. The use of rating scales in observational studies is not only time efficient but also gives a clinical picture which results in more specific implications for intervention purposes.

Another limitation of these studies is that generic parent-child interactions were not observed [2, 16-19]. It might be that diabetes-specific parent-child interactions affects child behavior (such as gaining independence,

stubbornness, oppositional behavior) and parental behavior (such as sensitivity, respecting autonomy, having fun together) [22] which might negatively affect the daily family life and the generic quality of the parent-child interaction.

The results of the cross-sectional studies of Patton et al. [2, 16, 17] showed that the behavior of the children and parents during mealtime were associated with the glycemic control (HbA_{1c}), the more family malfunctioning, the higher the glycemic control (HbA_{1c}). However, longitudinal studies are currently lacking in this area.

We expect that the way in which parents treating their child with T1DM during these often annoying, sometimes even invasive, yet unavoidable procedures can affect blood glucose control (HbA_{1c}) and psychosocial functioning. For example, if the mother responds anxiously when she has to monitor her child's blood glucose, the child may start to cry. If the mother then decides to postpone the finger prick this could lead to a hyperglycemia. Some parents may also have strong worries about future complications or hypoglycemic events. Because of these concerns, parents may decide monitor the child's blood glucose 15 times a day, three times per night, once at 23:00, and even once at 2:00 pm and once at 5:00 pm. These examples of non-constructive interaction patterns can be physically and emotionally stressful for the child (and parents), which might disturb the balance between effective treatment and optimal quality of life.

Because diabetes-related behaviors are usually consolidated in the first years post diagnosis [12, 23], interventions should start as early as possible. The combination of observing both generic and disease-specific interactions will identify interactional patterns to evaluate future behavioral interventions, with the aim of learning more effective parent-child interactions to optimize the glycemic control and psychosocial functioning of young children with T1DM as early as possible to prevent future problems.

In the past decades, several global rating scales have been developed for assessing different aspects of the quality of parent-child interactions, e.g., the Emotional Availability Scales (EAS) [24] and the scales developed by Erickson, Sroufe and Egeland [25]. These measures were designed to cover different aspects of parent-child interaction irrespective of an underlying disease. Disease-specific measures could assess interactions between parents and children at a disease-specific level (e.g., during administering insulin, mealtime behavior). Disease-specific instruments are expected to be more responsive to small changes that are important to clinicians or patients [26]. However, such a disease-specific observational measure is not available for children with T1DM. The main aim of the present study is to develop a disease-specific observational method, including a scoring system, to assess diabetes-specific parent-child interaction and to test the initial and preliminary psychometric properties of the pilot version

of the instrument. Additional aim is to explore whether the quality of parent-child interaction is associated with the glycemic control, child behaviors, and quality of life of children with T1DM.

Methods / Design

Developing the OKI-DO observation method

The development of the OKI-DO observation method (OKI-DO, **O**uder-**K**ind **I**nteractie **D**iabetes **O**nderzoek, which means: Parent-Child Interaction Diabetes Research) proceeds in two steps: (1) a small scale pilot study, and (2) a large scale validation study (see Figure 1).

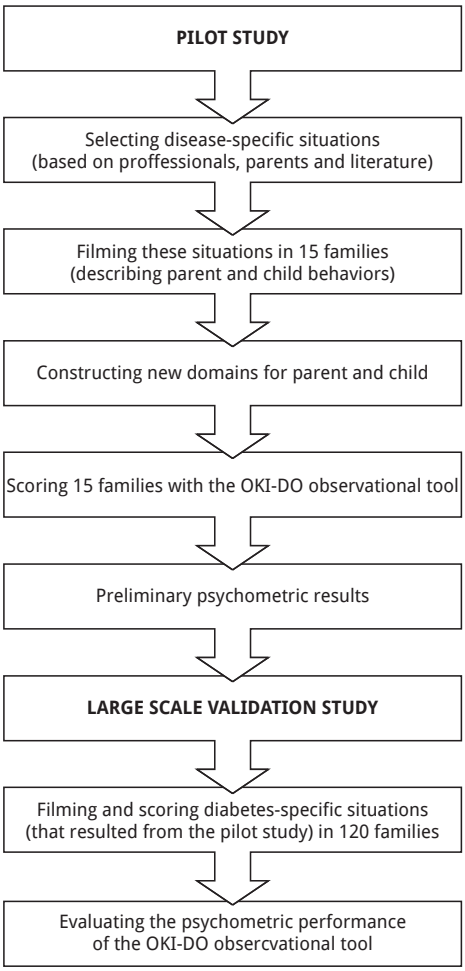

Figure 1: Development of the observational method to assess the diabetes-specific quality of the parent-child interaction: this figure shows the steps in developing and validating the OKI-DO method.

1. In the small scale pilot study we will develop the OKI-DO observation method in three steps: (a) defining relevant disease-specific situations; (b) development of a child and parent behavior list and making the observation method; (c) pilot test of the observational method. These three steps will be described below.

   a) The first step in the development of the OKI-DO method is to select disease-specific situations (e.g., administering insulin) that are most relevant for parents and children with T1DM. The selection of the situations will be based on theory, literature, and interviews with pediatricians, diabetes-nurses and parents.

   b) In the second step we will identify which disease-specific parent-child interactions play a role in diabetes-specific situations and how these interactions can be used to make our observation instrument. Therefore, the selected disease-specific situations will be videotaped during a home-visit in 15 families. Parent behaviors (e.g., comforting or distracting during the administering of insulin) and child behaviors (e.g., crying or accepting) will be described by a trained psychologist (HJAvB) and a research assistant (AMN). All parent and child behaviors will be described and will be specified from the observed parent and child behaviors by expert opinions, psychological theory, and the generic domains of parent-child interaction [24, 25]. The final result will be a list of rating scales, where each scale will be described by characteristics of parent-child interaction behaviors. We intend to develop complete descriptions of behaviors. An example of a pediatric diabetes-specific parent scale during administering insulin could be "parents supportive presence during administering insulin", which can be rated on a 7-point Likert scale varying from 1: "*Mother (or father) completely fails to be supportive to the child, being unavailable or being hostile toward the child when the child shows need of some support during this situation*" to 7: "*Mother (or father) skillfully provides support throughout the session. From the beginning she/he is confident that the child is capable of enduring this situation. If the child is having difficulty, she/he finds ways to calm the child and encourages positive behavior. Mother/father is not only emotionally supportive but continuously reinforces the child for good behavior (e.g., being compliant)*". In the

same manner other scales could be constructed, such as "*Parents respect for child's autonomy during administering insulin*" and "*Mothers/Fathers quality of instructions during administering insulin*". Scales of child behaviors might become for example "*Avoidance of the parent during administering insulin*" and "*Compliant/cooperative during administering insulin*".

c) The third and final step is to test the feasibility of the behavioral rating scale that has been developed in the previous two steps. The observational tool will be tested in a small-scale pilot study (N=15).

2. Second, we will apply the observational tool in a large scale study to collect data for further evaluation of the reliability and the validity of the instrument (N≈120). In addition, we will explore whether the quality of the parent-child interactions is associated with the glycemic control and psychosocial functioning (resilience, behavioral problems, and quality of life). Therefore, the parents are asked twice (with two years in between) to fill out questionnaires about psychosocial functioning of their child with T1DM. Glycemic control (HbA_{1c}) will be obtained from their medical records.

Procedure and participants

For this study, infants, toddlers and (pre)school children (aged 0-7 years) with T1DM and their parents will be recruited from several hospitals/institutions in the middle and southern part of the Netherlands (St. Elisabeth Hospital Tilburg, TweeSteden Hospital Tilburg, Catharina Hospital Eindhoven, St. Anna Hospital Geldrop, Bernhoven Hospital Veghel/Oss, Jeroen Bosch Hospital Den Bosch, Elkerliek Hospital Helmond, and Diabeter Rotterdam). We expect to approach about 175 young patients with type 1 diabetes mellitus and their parents. Assuming a response rate of 70%, we expect to collect data on 122 dyads in the present study. Families who agree to participate will be visited in their homes. After making an appointment, a set of questionnaires will be sent to the parents. The child and his or her parent(s) will be videotaped during 1. a free play task (e.g., playing with clay or making a puzzle), and 2. a number of disease-specific situations (e.g., administering insulin and mealtime behavior). The free play task will be rated with the scales developed by Erickson, Sroufe and Egeland [25], and the disease-specific situations will be rated with the OKI-DO instrument. At the end of the visit the questionnaires, which were filled out by the parents, will be collected. Glycemic control (HbA_{1c}) will be obtained from the medical record of the children.

Ethical considerations

The study design has been approved by the medical ethical committee of St. Elisabeth Hospital Tilburg (date: 25-05-2010). All parents/guardians are provided with written information about the study and are asked to give written informed consent prior to filming.

Study measures

Sociodemographic and clinical data

Parents will be asked to fill in a questionnaire with demographic background information (gender, date of birth, living situation, siblings, school, religion, land of birth, marital status, and education level) and clinical data (time since diagnose, disease-duration, treatment regimen, i.e., insulin pump, insulin injections, number of injections per day, number of glucose monitoring per day, number of hypoglycemic events for the past 3 months, hospital/institution, and hospitalization).

Quality of the parent-child interaction

In the large scale validation study, the disease-specific quality of the parent-child interaction will be measured with the observation method that has been developed in the small scale pilot-study (OKI-DO instrument). During a home-visit the child and his or her parents will be videotaped during 2 or 3 disease-specific situations (e.g., administering insulin). Their behaviors will be scored using the OKI-DO rating scales.

To assess generic quality of the parent-child interaction, the child and his or her parents are videotaped during a free play situation (e.g., playing with clay or making a puzzle; toys appropriate to the age and interests of the child) in the home situation, which will be rated by the scales developed by Erickson et al. [25]. These scales assess different domains of parent behavior and child behavior. Parental behavior includes the domains: supportive presence or the provision of emotional support, respect for the child's autonomy or non-intrusiveness, structure and limit setting, quality of instructions, and hostility. Child domains include negativity or anger, dislike or hostility, avoidance of interaction with the parent and compliance with suggestions and directions given by the parent. Each domain is scored on a scale ranging from 1 (low quality of parent-child interaction) through 7 (high quality of parent-child interaction).

Psychosocial functioning and quality of life of the children

Generic quality of life will be measured with the TNO-AZL Preschool Quality Of Life questionnaire or TAPQOL [27] in children in the age of 1 through 5 years of age and the TNO-AZL Child Quality Of Life questionnaire (TACQOL)

[28] in children of 6 years and older. These questionnaires measure parent's perceptions of health-related quality of life in (preschool) children. The TAPQOL is a multidimensional instrument with 43 items divided into 12 scales covering the following aspects: sleeping problems, appetite, lung problems, stomach problems, skin problems, motor functioning, social functioning, problem behavior, communication, anxiety, positive mood, and liveliness. The TACQOL is a multidimensional instrument with 63 items constituting 7 scales covering aspects of quality of life: five health-related scales: pain and symptoms, motor function, autonomy, cognitive functioning, interaction with parents and peers, and two scales that represent positive or negative emotions of a patient: experience of positive emotions and experience of negative emotions. In each of the health-related functioning scales, the parent can indicate to what extent specific problems occurred in the past few weeks, with three response categories: 'never', 'sometimes', and 'often'. If a problem occurs, the parents are asked how the child is feeling: '(very) good', 'not so good', 'pretty bad' and 'bad'. For each item, the two answers are combined into a single item score ranging from 0 to 4 ('never' 4 and 'sometimes' or 'often' combined with '(very) good' 3, 'not so good' 2, 'pretty bad' 1, and 'bad' 0). With the emotion scales, the parents indicate on a Likert scale or a certain emotion in their child has appeared in the last few weeks ('never', 'sometimes', 'often'). Item scores for the two emotion scales run from 0 to 2. In all TAPQOL and TACQOL scales, higher scores will correspond to a better quality of life. Reference values are given in the manual.

The diabetes-specific quality of life is measured with a child self-report questionnaire, composed by the Hvidøre Study Group. This questionnaire has been modified with permission of the authors so that the parents can complete the questionnaire for their child (proxy-report). The scale comprises 19 items about feelings of the child in relation with their diabetes (e.g. about administer insulin (injecting or pump) my child feels...), health (e.g. my child felt fit and healthy), leisure time (e.g. my child had enough time to play) and school (e.g. school / nursery / daycare went well). The items are rated on a 5-point Likert scale (e.g. 'very happy', 'happy', 'neutral', 'sad', and 'very sad'). Scores are coded so that a higher score corresponds to a better quality of life. Currently, this questionnaire is widely used, and will be validated, by the Hvidøre Study Group.

The degree of psychosocial problems will be measured using the Strengths and Difficulties Questionnaire (SDQ) [29]. The SDQ is a brief behavioral screening questionnaire and measures the presence of psychosocial problems, the strengths of the child and the influence of psychosocial problems in daily functioning. The SDQ is suitable for children of 3 years and older. The questionnaire contains 25 items, covering the following five domains: conduct symptoms, hyperactivity/ inattention, emotional problems, peer relationship

problems, and pro-social behavior. The 25 items were formulated on the basis of propositions (e.g., "Considerate of other people's feelings") and relate to the past 6 months. Some propositions are oppositely formulated, such as "Thinks before acting out." Therefore, the subscales have a bipolar character, that is, a low score not only means that there are no problems, but also that there are one or more strengths [30]. Research showed that the SDQ is a reliable and valid questionnaire [31].

To measure potential behavioral problems of children less than three years, the Child Behavior Check List (CBCL) [32] is used. The CBCL 1.5-5 consists of 99 items that are categorized into seven scales, including emotionally reactive, anxious / depressed, somatic complaints, withdrawal, sleep problems, attention problems, and aggressive behavior. The items can be further summarized into internalizing problems and externalizing problems or a total problem score (adding up all items).

Data analyses

The aim of this study is to develop an observational method that provides a standardized procedure to collect information on parent-child interactions in young children by means of direct observations from videotaped diabetes-specific situations. Before the observational method can be used for substantive research purposes, it has to be empirically examined to what extent the observation method provides reliable and valid information on diabetes-specific parent-child interactions. In this study, we will first test the observational tool in a small-scale pilot study (N=15). Second, we will apply the observational tool on a larger scale to collect data for further evaluation of the reliability and the validity of the instrument (N≈120).

1. Small scale Pilot Test.

In the pilot study, three raters will score parent-child interactions for 15 pediatric diabetes patients in videotaped home situations. Two of the project leaders (EEH, HJAvB) and a research assistant (AMN) will independently rate the video-tapes. One of the raters (HJAvB) is an expert on parent-child interactions and has a lot of experience with various rating scales in different populations. After the raters have rated the 15 patients, a debriefing questionnaire will be administered in which the raters are asked to appraise the feasibility of the observational tool (are the instructions clear; do instructions need further explications) and to comment on usefulness and face validity of the scales and its constituent indicators. In addition, the inter-rater reliability will be determined by means of inter-rater reliability indices (e.g., Intra Class Correlation Coefficients, ICC). Based on qualitative data from the rater's feedback and the statistical

result, the observational tool may be refined if necessary (e.g., making the instructions more specific or explicit). If major revisions are needed, the pilot test will be repeated using same groups of raters. The pilot study must result in an experimental version of the observational tool that is deemed applicable, valid, and appropriate for diabetes-specific situations by an expert panel and the raters.

2. Large Scale Validation Study.

In the validation study, data of 120 (pre-)school children with T1DM and their parents from several hospitals/institutions will be analyzed. The reliability of the OKI-DO observation method is defined by the degree of inter-rater reliability and the reliability of average ratings across raters, inter-rater agreement, and will be assessed using weighted kappa [33] and intra-class correlation coefficients [34]. Reliability values ≥ 0.70 are generally accepted as adequate for scientific research. The validity of the OKI-DO observation method is assessed in different ways.

First, we will evaluate the face validity of the instrument. We will describe all observed parent and child behaviors as complete and precisely as possible to make sure the OKI-DO observation method will measure the quality of interaction between parent and child.

Second, we will examine construct validity by testing predictions on the relations with other parent-child interaction scales. If these predictions are supported by the data, we have supportive evidence for the construct validity of the instrument [35]. In particular, we will test associations of scores obtained with the OKI-DO method with scores from related observations tools assessing global parent-child interaction [25]. We expect that global interaction scales [25] and the OKI-DO scales will correlate substantially (Spearman's rho > 0.40), whereas conceptually unrelated scales (like 'negativity' and 'enthusiasm') will correlate less than 0.20. We will also compare the correlation of a few scales of the included questionnaires with the OKI-DO method. One example includes a comparison of the scale 'aggressive behavior' from the CBCL with a scale from the OKI-DO method that measures something like 'hostility of the child'.

Third, validity will be assessed with the method of known-group comparisons [36] to evaluate the extent to which the observational tool will be able to discriminate between subgroups of patients differing in time since diagnosis and glycemic control ('good' versus 'bad' HbA_{1c}) and subgroups of parents differing in gender (father/mother). We believe that children who are recently diagnosed with T1DM and children who have a 'bad' glycemic control will have a poorer quality of parent-child interaction than children who are diagnosed with T1DM for a long time and children with a 'good' glycemic control. We also believe that mothers and fathers will differ in the quality of parent-child interaction, because mothers

mostly have a closer relationship with their child [37].

3. Substantive analysis on relation between parent-child interactions, psychosocial functioning and glycemic control.

Third, we will study the relationship between parent-child interactions as measured by our observational tool and with the generic scales developed by Erickson et al. [25] with (changes in) the glycemic control (HbA_{1c}), and psychosocial functioning (resilience, behavioral problems, and quality of life) of the children. These relationships will be tested with multiple regression analyses.

Sample size / power analysis

We expect to include about 120 families (parent-child pairs) in this study, in which all 120 pairs are judged by three raters. To justify the sample size, we did several checks on the precision with which inter-rater reliability can be estimated and the power of testing hypotheses on correlations.
For assessing the inter-rater reliability (i.e., consistency among fixed raters), we use the ICC [34]. Using confidence intervals reported in Shrout and Fleiss [34] we can assess the precision of ICC estimates. For 120 observations and three raters, a 90% CI for an ICC of .70 ranges from .63 to .76. Furthermore, with 120 observations and three raters, we have 80% power to find an inter-rater reliability of .74 or higher when tested against .70 (two-tailed test; α = .05). For the substantive analyses (i.e., testing relations of ratings with other variables), we will use mean ratings across the three raters per parent-child dyad. With 120 observations and three raters, a 90% confidence interval for a reliability of .80 for the mean ratings runs from .74 to .85.

To estimate the inter-rater agreement, which reveals important information about the feasibility of the instrument as an observational tool in clinical practice, we examine pair wise inter-rater agreement using kappa (i.e., treating observations as nominal ratings) and weighted kappa (treating observations as ordinal ratings) [38]. Using results from Hanley [39], the expected standard error of kappa in a sample of 120 will be in between .07 (for kappa 0.3) and .05 (for kappa = .8). For example, for a kappa value of .6, the 90% confidence interval ranges from .48 to .72. This approximation is based on 4-point ratings. However, as we will use seven-category ratings, the true standard error will likely be smaller and our kappa estimates will be more precise than indicated here. For valid application of weighted kappa for ratings on 7-point Likert scales, we used results from Cicchetti [40], who showed that the required minimum sample size is given by $2 \times 7^2=98$.

Power with respect to correlations: With Gpower 3.0 we calculated that finding population correlations of .4 or higher with at least a power of .9 (two-

tailed t-test; α = .05) minimum samples sizes of 58 are needed. Power analyses showed that with regression analyses there will be sufficient power to find an explained variance of 15 percent or more (F-test, α = .05). For example, for 4 predictors and N = 120, the power for finding effect sizes of magnitude $R2 \geq .15$ is .93 (calculated with Gpower 3.0).

In sum, power and precision analysis showed that 120 dyads should be suffice to accurately estimate inter-rate reliability and agreement, and in sufficient power to find correlations between measures of parent-child interactions and the outcome measures.

Possible results / relevance

The availability of a disease-specific observational instrument will enable the detailed assessment of the quality of the disease-specific parent-child interaction. More specifically, the instrument can be used to conduct studies that can help to determine which parent-child interaction-patterns are associated with specific diabetes outcomes, such as self-care and glycemic control (HbA_{1c}). Results will also show whether, and how, quality of the disease-specific interaction during these procedures is related to generic interaction between children and their parents. Further, we expect that the quality of the parent-child interaction will appear to affect glycemic control (HbA_{1c}), child behaviors and quality of life. In future research, the OKI-DO observational method can be used as an evaluative tool to measure changes in the diabetes-specific interaction patterns across time, e.g., as a result of interventions based on the outcomes of this study.

Discussion

The purpose of this study is to develop an observational tool to assess the disease-specific parent-child interactions in families with young children with T1DM. We will test initial and preliminary psychometrics of the pilot version of the OKI-DO instrument and then test the psychometric performance of the OKI-DO instrument in the large scale validation study. Sound observational methods enable scientists and clinical practitioners to compare objective behaviors at different time points and to evaluate interventions. When the OKI-DO method appears to be psychometrically sound, it will be used in future studies. The observational tool will be made available for (international) use by other research groups as well (English version).

To minimize observational bias, we will observe parent-child interactions in the home situation, where it is more likely that routine and daily patterns of interaction will be revealed compared to clinical settings [41].

Results of the present disease-specific observation study will identify behaviors that should be targets for future behavioral interventions for young

children with T1DM and their parents. For health care providers to be able to inform, treat and refer patients and their parents to needed types of care, it is important to have insight into the parent (mothers and father) –child interactions in families with these young patients. Moreover, as diabetes-related family behaviors seem to be established in the early years post-diagnosis, interventions should start as early as possible. Ideally, future interventions should increase the strengths and decrease the weaknesses of global and disease-specific interaction patterns in families with children with T1DM, preferably in early childhood. Or, as an English proverb says "*What's learnt in the cradle lasts till the tomb*".

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Chapter 4

Qualitative observation instrument to measure the quality of parent-child interactions in young children with type 1 diabetes mellitus

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ABSTRACT

Introduction
In young children with type 1 diabetes mellitus (T1DM), parents have complete responsibility for the diabetes-management. In toddlers and (pre)schoolers, the tasks needed to achieve optimal blood glucose control may interfere with normal developmental processes and could negatively affect the quality of parent-child interaction. Several observational instruments are available to measure the quality of the parent-child interaction. However, no observational instrument for diabetes-specific situations is available. Therefore, the aim of the present study was to develop a qualitative observation instrument, to be able to assess parent-child interaction during diabetes-specific situations.

Methods
First, in a pilot study (n=15), the observation instrument was developed in four steps: (a) defining relevant diabetes-specific situations; (b) videotaping these situations; (c) describing all behaviors in a qualitative observation instrument; (d) evaluating usability and reliability. Next, we examined preliminary validity (total n=77) by testing hypotheses about correlations between the observation instrument for diabetes-specific situations, a generic observation instrument and a behavioral questionnaire.

Results
The observation instrument to assess parent-child interaction during diabetes-specific situations, which consists of ten domains: "emotional involvement", "limit setting", "respect for autonomy", "quality of instruction", "negative behavior", "avoidance", "cooperative behavior", "child's response to injection", "emphasis on diabetes", and "mealtime structure", was developed for use during a mealtime situation (including glucose monitoring and insulin administration).

Conclusions
The present study showed encouraging indications for the usability and inter-rater reliability (weighted kappa was 0.73) of the qualitative observation instrument. Furthermore, promising indications for the preliminary validity of the observation instrument for diabetes-specific situations were found (r ranged between |.24| and |.45| for significant correlations and between |.10| and |.23| for non-significant trends). This observation instrument could be used in future research to (a) test whether parent-child interactions are associated with outcomes (like HbA_{1c} levels and psychosocial functioning), and (b) evaluate interventions, aimed at optimizing the quality of parent-child interactions in families with a young child with T1DM.

Introduction

When young children are diagnosed with type 1 diabetes mellitus (T1DM), parents have to take complete responsibility for the daily diabetes-management of their child, 24 hours a day, 7 days a week. Several times a day, they have to monitor the blood glucose level, administer insulin, regulate food intake, and guard these parameters in conjunction with the level of physical activity of their child with diabetes. The tasks needed to achieve optimal blood glucose control, however, may interfere with normal and age appropriate behaviors that occur in the toddler and pre-school years (e.g., increase in autonomy, independence-seeking, refusing food, or oppositional behavior) [1]. This may also affect the quality of the parent-child interaction [2]. If the quality of the parent-child interaction is non-optimal, this might also have a negative impact on the diabetes self-care behaviors of parents and children.

Given the importance of this topic, it is surprising that the number of studies examining the quality of the parent-child interaction in families with young children with T1DM is limited [3]. Few studies have examined the quality of parent-child interaction in children with T1DM, and existing studies mainly focused on older children with T1DM (> 8 years) [4-8] or used a wide age range, from 1-14 years [9, 10]. Moreover, most studies mainly used self-report measures or semi-structured interviews [4, 5, 7, 9-11]. When investigating the quality of parent-child interaction, videotaped interactions and rating scales may give more detailed information about the various aspects of parent-child interactions [12, 13]. Moreover, self-report measures and interviews by definition reflect a subjective quantification of concepts from the perspective of parents whereas observed interactions by an independent observer can provide more objective data [3].

As far as we know, only two research groups were identified that studied the quality of parent-child interaction in young children with T1DM using an observation method [1, 14-18]. These studies observed behaviors of both parents and children during mealtime, which were counted by frequency (e.g., how often the child was encouraged by the parents to keep eating), time intervals (e.g., see if the child is eating on second 10, second 20 etc.) or whether a specific behavior was present or not during the observation. However, counting the number of specific behaviors and using time intervals have some disadvantages. No information about the affective quality of the dyadic behavior is reflected in the observed behaviors. In contrast, applying rating scales has the advantage that affective components can be taken into account. This way of coding observational data allows making many dimensions and subtle differences in behaviors. Moreover, the predictive value of rating scales has proved to be more accurate than just counting specific behaviors [19-21]. Furthermore, when using

rating scales, the behaviors of the parents can be evaluated in the context of the behaviors of the child [20], which is important when observing the quality of parent-child interaction. An additional advantage of rating scales is that it is more time efficient [12], and can cost up to five times less time than counting all behaviors [19]. Hence, the use of rating scales in observational studies gives a clinical picture which results in more specific implications for intervention purposes and is time efficient [3].

In the past decades, several generic rating scales have been developed for assessing different aspects of the quality of parent-child interactions for use in the general population [3], e.g., the Emotional Availability Scales (EAS) [22], scales developed by the National Institute of Child Health and Human Development (NICHD) [23], and scales developed by Erickson, Sroufe and Egeland [24]. These rating scales were designed to cover various aspects of parent-child interaction irrespective of an underlying disease. Moreover, these tools were not specifically designed to use in a clinical sample and did not take into account disease-specific behaviors such as the parent's and child's reaction to medical tasks or the emphasis of the disease during the interaction. However, disease-specific observational rating scales are still not available.

The aim of this study is therefore to develop such an observation instrument for diabetes-specific situations to assess the quality of parent-child interactions in young children (0-7 years) with T1DM by means of direct observations. Furthermore, the usability, inter-rater reliability and preliminary validity of the observation instrument are investigated.

Methods

Participants and procedure

At first, all infants, toddlers and (pre)school children (aged 0-7 years) treated for T1DM and their parents were recruited from Kidz&Ko, a partnership between seven pediatric diabetes clinics, and Diabeter, a national center for pediatric and adolescent diabetes care and research. Due to a small sample size, we also recruited all children (0-7 years) with T1DM from 7 other hospitals in the Netherlands (Isala Clinics Zwolle, Amphia Hospital Breda, Franciscus Hospital Roosendaal, Academic Hospital Maastricht, Medical Spectrum Twente Enschede, Zorg Groep Twente Almelo/Hengelo, Atrium Medical Center Heerlen). In these 15 hospitals, 138 young children with T1DM were treated. Parents who lacked basic proficiency in Dutch were excluded, as well as children who were mentally disabled and/or had Down syndrome, or were diagnosed with an Autism Spectrum Disorder (total families excluded: n=17). Of the 121 eligible parents of children with T1DM, 77 families (64%) agreed to participate. Reasons for not participating were: not willing to be videotaped (n=18), a recent hospitalization of

the child (n=3), loss of a family member (n=1) or personal reasons (n=22).

During a home-visit, a diabetes-specific situation (i.e., mealtime observation) and a free play situation were videotaped. During the structured 10-minute free play situation (playing with clay or making a puzzle) one of the parents and their child were asked to play together as they normally would do (this parent was also the focus of the parent-child interaction during the diabetes-specific situation). Furthermore, parents were asked to fill out a questionnaire with (socio)demographic characteristics (i.e., gender of the child, age of the child, marital status parents, and educational levels of both parents) and clinical characteristics (i.e., treatment regimen, times they monitored their child's blood glucose level a day (average), and years since diagnosis), specifically designed for this study. Glycosylated hemoglobin (HbA_{1c}), measured closest to the home-visit, was locally determined at the hospital the child was treated and extracted from the medical record. Furthermore, to examine preliminary validity of the observation instrument for diabetes-specific situations, we asked the parents about their children's behavior by filling out the Strengths and Difficulties Questionnaire (SDQ) [25]. When examining the preliminary validity of the observation instrument, we only used the data of SDQ questionnaires of the parent that was the focus during the videotaped situation. The SDQ is a brief behavioral screening questionnaire and measures the presence of psychosocial problems and the strengths of the child. The questionnaire consists of 25 items, covering the following five domains: emotional problems, conduct problems, hyperactivity/inattention, peer relationship problems, and pro-social behavior. The 25 items are formulated on the basis of propositions and relate to the past 6 months. Some propositions are oppositely formulated. Therefore, the subscales have a bipolar character: a low score not only means that there are few problems, but also that there are strengths [25]. Research showed that the Dutch translation of the SDQ has acceptable to good psychometric properties [26].

The study was approved by the Medical Ethical Review board of St. Elisabeth Hospital Tilburg (date: 25-05-2010) and in conjunction with the Helsinki Declaration on human research.

Constructing the observational rating scales

The development of the observation instrument has proceeded in (1) a pilot study in which we developed and evaluated the observation instrument, and (2) a subsequent study to assess preliminary validity of the observation instrument for diabetes-specific situations [3].

1. In the pilot study (n=15) we developed the observation instrument in four steps (see Figure 1):

The first step was to determine diabetes-specific situations that were most salient and/or problematic for parents and children with T1DM. The selection of these situations was based on literature and interviews with four pediatricians, four diabetes-nurses and four randomly selected parents (parents who were in the waiting room after our visit to the pediatrician) in which they were asked: "I*n which situations parents might encounter problems with the diabetes-management and behavior of their child?*"

In the second step we videotaped the most salient diabetes-specific situation and a generic situation (free play) during a two-hour home-visit in 15 families after receiving written consent from the parents. The observer did not participate in the family interactions during the diabetes-specific situation and free play situation. Because the observer did not participate and kept herself aloof from the situation, no observer effects are expected. The behaviors recorded during the free play situations were scored with an observation instrument to assess generic parent-child interaction, developed by Erickson, Sroufe and Egeland [24]. This generic observation instrument consist of six parent domains ("Supportive presence", "Respect for child's autonomy", "Structure and limit setting", "Quality of instruction", "Hostility", and "Confidence"), and eight child domains ("Negativity", "Avoidance of parent", "Compliance/child complies with parent's task direction", "Affection toward parent", "Persistence", "Reliance on parent for help", "Enthusiasm", and "Experience of the session"). At the end of each home-visit, parents were asked if the videotaped situations were typical or different from other days (for example, if they and/or their children did behave more active or more withdrawn than they normally do).

The third step was to describe all videotaped behaviors in an observation instrument for diabetes-specific situations (based on generic observation instruments [22-24], but specifically described to assess the quality of parent-child interaction during diabetes-specific situations).

The fourth and final step of the pilot study was to test the usability and the inter-rater reliability of the developed observation instrument. Results of the pilot study were used to refine the observation instrument for diabetes-specific situations.

2. Second, we conducted the same home-visit as in the pilot study in 62 additional families (total sample n=77), to collect data for preliminary validity of the observation instrument.

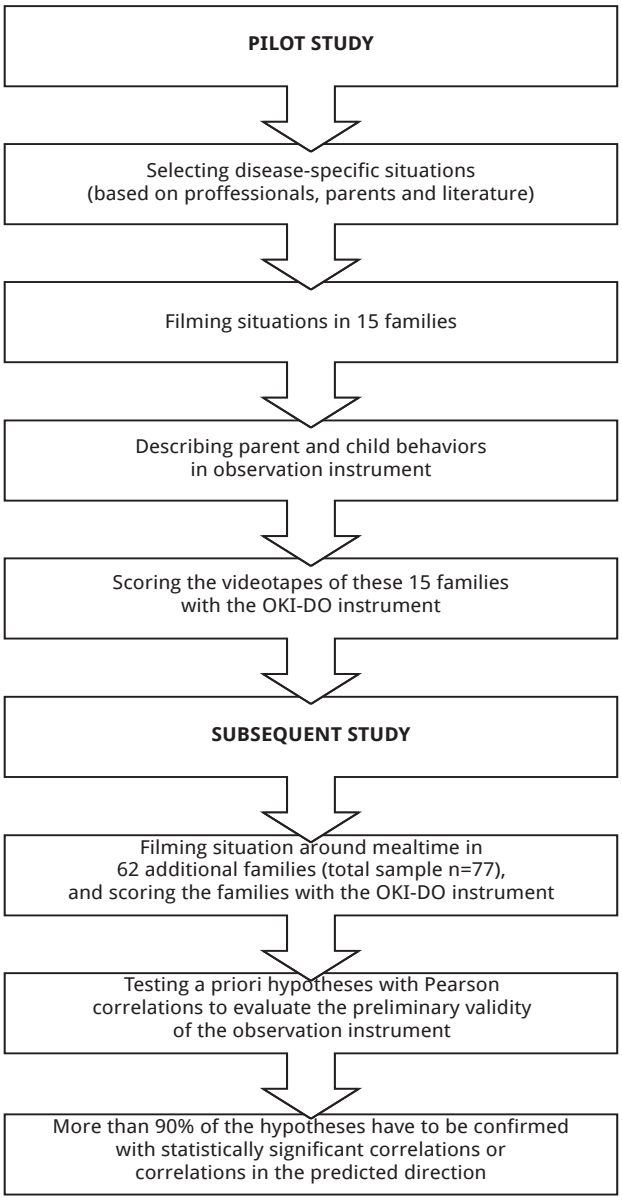

Figure 1: Constructing the OKI-DO observation instrument

Statistical analyses

In the pilot study, the usability of the observation instrument was determined by a debriefing questionnaire in which the raters and authors were asked to appraise the usability of the observation instrument (Were the instructions clear?; Did instructions need further explications?; Were the

descriptions of behaviors complete?; Were all behaviors covered?). To determine the inter-rater reliability [27], two raters (HvB and AN), with previous experience in observing and rating behaviors with generic observation instruments, scored the first 15 videotapes independently. A weighted kappa between 0.61-0.80 is generally regarded as an indication of substantial agreement [27].

The validity of observational rating scales is often assumed and not examined [28], however, we did investigate preliminary validity of the observation instrument in a subsequent study. The preliminary validity was evaluated by testing a priori hypotheses on the association between the observation instrument for diabetes-specific situations (observation during a diabetes-specific situation) and an observation instrument to assess generic parent-child interaction[24] (observation during free play). The hypotheses were based on the model of Belsky [29], which encompasses three determinants that influence parenting: characteristics of the parent, characteristics of the child, and contextual sources of stress (in this case T1DM) and support. For example: children, who show lots of affection toward their parent, probably contribute to a better quality of parent-child interaction. Or parents, who are hostile toward their child, probably contribute to a lower quality of parent-child interaction. Also, as youth (11-18 years) with T1DM with a disturbed parent-child relationship show more behavioral problems [6], a priori hypotheses between the domains of the observation instrument for diabetes-specific situations and SDQ subscales or total score were formulated to further examine the preliminary validity of the observation instrument. At least 75% of the hypotheses must be confirmed by a correlation (significant or non-significant trend) in order to demonstrate the preliminary validity of the observation instrument [30]. Furthermore, effects sizes will also be examined. According to Cohen, r of 0.1, 0.3, and 0.5 can be considered as small, medium and large effects, respectively [31].

Results

Participants

Table 1 summarizes the characteristics of participating parents and children. Among the participating children, there were 41 boys (53%). The children with T1DM had a mean age of 5.12 years (SD=1.52, range: 2-7 years). Most children (82%) received pump therapy. On average, parents monitored their child's blood glucose 6 times a day (range: 2-20). The mean HbA_{1c} value of the children was 59 mmol/mol or 7.6% (range 32-80 mmol/mol or 5.1% - 9.5%). Of the 74 mothers and 3 fathers that were observed, 67 mothers (91%) and 3 fathers (100 %) completed the form with the (socio)demographic characteristics and SDQ [25]. Most mothers (83%) and fathers (100%) were cohabiting or married/ registered partners (7% of the mothers and 0% of the fathers were single). Half of

the participating mothers (50%) had a higher educational level (i.e., approximately 12 years of formal education), while all fathers (100%) had a Bachelor's or Master's degree (i.e., approximately 15 years of formal education).

Table 1: Sociodemographic and clinical characteristics of young children with type 1 diabetes and their parents.

			N (%)	M	SD
Children	Sex	Boys	41 (53%)		
		Girls	36 (47%)		
	Age (years)	(range 2-7)		5.12	1.52
	HbA_{1c}	(range 32mmol/mol-80mmol/mol)	77	59.25	9.03
		(range 5,1% - 9,5%)	77	7.6%	0.8%
	Treatment	Insulin pump	63 (82%)		
		Multiple daily insulin injections	14 (18%)		
	Blood glucose monitoring	Times a day (range 2-20)		6.08	2.69
	Years since diagnose	(range 1-6 years)		2.61	1.44
Parents	Total	Mothers observed	74 (96%)		
		Fathers observed	3 (4%)		
	Marital status (mothers)	Single	5 (7%)		
		Cohabiting with partner	10 (13%)		
		Married / registered partners	52 (70%)		
		Missing	7 (10%)		
	Marital status (fathers)	Single	0 (0%)		
		Cohabiting with partner	1 (33%)		
		Married / registered partners	2 (67%)		
		Missing	0 (0%)		
	Educational level (mothers)	Primary education	1 (1%)		
		About 12 years of formal education	36 (49%)		
		15-16 years of formal education	29 (39%)		
		Other	1 (1%)		
		Missing	7 (10%)		
	Educational level (fathers)	Primary education	0 (0%)		
		About 12 years of formal education	0 (0%)		
		15-16 years of formal education	3 (100%)		
		Other	0 (0%)		
		Missing	0 (0%)		

Pilot study

When interviewing the experts about which situation to observe, glucose monitoring and the mealtime were both mentioned ten times. Nighttime (because of possible nocturnal hypoglycemia), unexpected situations (such as unexpected treats) and diabetes-management at school were mentioned nine, seven and one time(s) respectively. These situations were confirmed by the literature [11, 15-18, 32-41]. Because unexpected daily situations, nighttime observations, and school observations are of course more difficult and impractical to record, and filming the night-time situation can be perceived as too intrusive, we refrained from using these situations in our study. Therefore, we decided to observe the mealtime situation (including glucose monitoring and insulin administration) as this was also most frequently mentioned by the experts. Because of work, daycare and/or school, we videotaped dinnertime with all siblings and both

parents present (only one parent was observed for scoring). If it was not possible to observe dinnertime, we videotaped lunchtime (with siblings, but only one parent present). On average, the mealtime lasted about 25 minutes.

After videotaping the mealtime situation (including glucose monitoring and insulin administration) in 15 families, parents of two families indicated that they or their children acted a bit different in the beginning of the home-visit when the camera was introduced (before the actual observation started), but also that after a while they themselves and their children did not even notice the camera. Based on the videotapes, the observer described all observed parent behaviors (e.g., being emotionally involved) and child behaviors (e.g., crying or accepting) in the observation instrument for diabetes-specific situations (based on generic observation instruments [22-24], but with a focus on the parent-child interaction related to the child's diabetes and diabetes-related tasks). The observed behaviors, together with expert views of two other observers, a pediatrician and a diabetes nurse, and generic domains of parent-child interaction [22-24], resulted in a qualitative observation instrument for scoring behavior during mealtime (including glucose monitoring and insulin administration) in young children with T1DM. The observation instrument was named **OKI-DO** (OKI-DO, which literally means **O**uder **K**ind **I**nteractie-**D**iabetes **O**nderzoek: Parent Child Interaction-Diabetes Research). The qualitative observation instrument comprises ten domains to assess the quality of the parent-child interaction in diabetes-specific situations, including four parent domains ("emotional involvement", "limit setting", "respect for autonomy", and "quality of instruction"), four child domains ("negative behavior", "avoidance", "cooperative behavior", and "child's response to injection"), and two family-domains ("emphasis on diabetes" and "mealtime structure"). All the domains consist of qualitative descriptions of the behavior or situation on a 5-point Likert scale. Higher scores reflect more of the behavior (e.g., a high score on 'emotional involvement' means the parent is highly emotional involved, and a high score on 'negative behavior' means the child shows a lot of negative behavior). An example of the domain "*respect for autonomy*" can be rated varying from score 1: '*The caregiver receives this score if he/she fully determines what should happen without explaining anything to the child and with a visible lack of respect for the autonomy. For example: the caregiver just takes the finger of the child to check the glucose, (harshly) 'pulls' the child in the correct position to operate the insulin pump or determines (without consulting or warning the child) where and when the insulin injection takes place, the caregiver fully determines what and how much the child eats. If the child is (rather) independent in managing his/her diabetes, the caregiver receives this score if he/she repeatedly interferes when the child is managing his/her diabetes, while it is clear from the observation that the child can perform everything on*

its own', to score 5: '*The caregiver receives this score if he/she praises initiatives of the child and encourages the child to make decisions on his own regarding his/her diabetes. The child may, for example, read the glucose meter, operate the insulin pump or determine where and when the insulin injection takes place (the caregiver could of course check the things his/her child does, but is herein not at all intrusive). Everything is determined in consultation with the child and the child is treated with respect*.' The parent-child dyad will receive one score (1-5) on all ten domains of the OKI-DO instrument. All domains, like "*respect for autonomy*" as described above, focus on the parent-child interaction related to the child's diabetes and diabetes-related tasks. Finally, we have written a manual with detailed instructions how to videotape, observe and score the quality of the parent-child interactions.

The last step of the pilot study was to test the usability and the inter-rater reliability of the concept version of the OKI-DO instrument. The responses on the debriefing questionnaire (see *Statistical analyses*) to determine the usability, yielded a few minor improvements. These improvements consisted of specifying the instructions and more detailed descriptions of specific behaviors. Two raters (HvB and AN) with experience in rating with generic observation instruments independently scored the videotapes of the 15 families with the improved OKI-DO instrument. Weighted kappa was 0.73, indicating a good inter-rater reliability [27].

To conclude, the pilot study resulted in an observation instrument that appeared to be usable and reliable to assess parent-child interaction during mealtime (including glucose monitoring and insulin administration) in families with a young child with T1DM.

Subsequent study

In the subsequent study, we scored all families with the OKI-DO instrument (total n=77). Table 2 shows the mean, minimum and maximum scores on the domains of the OKI-DO instrument. As Table 2 shows, the current sample consists of rather high-functioning families (low scores on '*negative behavior*', '*avoidance*', '*response to injection*', and high scores on '*limit setting*', '*respect for autonomy*', '*cooperative behavior*', which means that the participating families in our study did not encounter major problems during mealtime, glucose monitoring and insulin administration).

Table 2: Mean, minimum and maximum scores on the OKI-DO domains.

OKI-DO domains	Mean	Minimum	Maximum
Emotional involvement	3,9	2	5
Limit setting	4,2	2	5
Respect for autonomy	4,1	2	5
Quality of instruction	3,5	1	5
Negative behavior	1,5	1	4
Avoidance	1,7	1	4
Cooperative behavior	4,1	2	5
Child's response to injection	1,7	1	4
Emphasis on diabetes	2,5	1	5
Mealtime structure	3,6	1	5

To investigate preliminary validity of the OKI-DO instrument, we tested Pearson correlation coefficients between the OKI-DO instrument (observations during mealtime, including glucose monitoring and insulin administration) and a generic observation instrument [24] (observation during free play). Also correlations between the OKI-DO instrument and SDQ subscales or total score were examined to further examine the preliminary validity of the OKI-DO instrument. Table 3 (highlighted in gray) and Table 4 show the a priori hypotheses of predicted correlations between the OKI-DO instrument and generic observation instrument to assess generic parent-child interaction [24] or the SDQ [26].

Table 3 shows the correlation coefficients between the OKI-DO instrument during mealtime (including glucose monitoring and insulin administration) and the generic observation instrument [24] during free play. As Table 3 shows, 32 out of 34 (94%) hypothesized correlations (highlighted in gray) between the OKI-DO instrument and the generic observation instrument [24] showed small to medium effect sizes [31] and were confirmed with 19 statistically significant correlations (range |0.24| to |0.45|) and 13 non-significant trends (range |0.10| to |0.23|, as a correlation of 0.24 was significant, we decided that correlations of |0.10| or higher were non-significant trends). This distribution is a positive indication for the preliminary validity of the OKI-DO instrument. The OKI-DO domain "*quality of instruction*", showed a zero correlation with the generic domains "*structure and limit setting*", and "*quality of instruction*", although we expected a (significant) correlation.

Furthermore, we found a few unpredicted significant correlations between the OKI-DO domains and generic domains [24]. However, these correlations are no evidence for or against the preliminary validity.

Table 3: Correlation coefficients between the OKI-DO instrument and generic observation instrument [24] during free play.

Free play (generic)	OKI-DO	*Parent* Emotional involvement	Limit setting	Respect for autonomy	Quality of instruction	*Child* Negative behavior	Avoidance	Cooperative behavior	Child's response to injection	*Family* Emphasis on diabetes	Mealtime structure
Parent's	Supportive presence	.38**	.22	.24*	.02	-.13	-.15	.16	.14	-.13	.16
	Respect for autonomy	.40**	.15	.31**	.20	-.12	-.22	.17	.03	-.01	.00
	Structure and limit setting	.18	.31**	.23	.05	-.20	-.14	.20	.01	-.10	.24*
	Quality of instruction	.17	.12	.09	.07	-.13	.01	.00	.03	-.05	.03
	Hostility	-.35**	-.11	-.45**	-.08	-.02	.28*	-.18	-.12	.16	-.02
	Confidence	.18	.35**	.14	-.04	-.28*	-.14	.23	.02	-.13	.38**
Child's	Negativity	-.19	-.15	-.16	-.12	.14	.08	-.14	-.10	.13	.01
	Avoidance of parent	-.09	-.13	-.19	-.08	.12	.30**	-.01	-.01	.08	-.02
	Compliance	.18	.26*	.33**	.21	-.10	-.15	.14	.14	-.09	.05
	Affection toward parent	.28*	.18	.33**	.07	-.17	-.13	.18	.16	-.03	.01
	Persistence	.16	.28*	.01	-.03	-.09	-.04	.19	.09	.22	.26*
	Reliance on parent for help	.15	-.09	-.10	-.00	.04	.07	-.07	.07	.02	.02
	Enthusiasm	.14	.12	.08	-.17	-.01	.02	.04	.18	.27*	-.06
	Experience of the session	.33**	.25*	.29*	.02	-.05	-.00	.12	.19	.07	.18

▭ = Expected correlation
* = Correlation is significant on a 0.05 level
** = Correlation is significant on a 0.01 level

Table 4 shows the correlation coefficients between the OKI-DO instrument and the SDQ [25] subscales or total score. All (100%) of the hypothesized correlations showed small to medium effect sizes [31] and were confirmed with statistically significant correlations or non-significant trends. This is a further positive indication for the preliminary validity of the OKI-DO instrument.

Table 4: Correlation coefficients between the OKI-DO observation instrument and SDQ [25] subscales or total score.

OKI-DO instrument	Expected direction:	SDQ scale:	Result:
Emotional involvement	+	Total problems	.10
Limit setting	-	Conduct problems	-.23
Respect for autonomy	+	Prosocial behavior	.11
Quality of instruction	-	Total problems	-.14
Negative behavior	+	Conduct problems	.20
Avoidance	-	Prosocial behavior	-.19
Cooperative behavior	-	Conduct problems	-.27*
Child's response to injection	xxx	xxx	xxx
Emphasis on diabetes	xxx	xxx	xxx
Mealtime structure	-	Conduct problems	- .11

* = Correlation is significant on a 0.05 level

To conclude, the present study showed encouraging indications for the usability, inter-rater reliability, and preliminary validity of the OKI-DO observation instrument to assess parent-child interaction in young children with T1DM during mealtime (including glucose monitoring and insulin administration).

Discussion

The purpose of the present study was to develop a qualitative observation instrument to assess parent-child interaction in young children (0-7 years) with T1DM in diabetes-specific situations. In a pilot study (n=15) we developed the OKI-DO observation instrument for scoring parent and child behavior during mealtime (including glucose monitoring and insulin administration), which consists of: "*emotional involvement*", "*limit setting*", "*respect for autonomy*", "*quality of instruction*", "*negative behavior*", "*avoidance*", "*cooperative behavior*", "*child's response to injection*", "*emphasis on diabetes*", and "*mealtime structure*". The OKI-DO instrument appeared to be suitable to assess parent-child interaction in diabetes-specific situations, as weighted kappa indicated a good inter-rater reliability and the subsequent study showed positive indications for the preliminary validity. We examined the preliminary validity of our instrument in the total sample (n=77) by testing hypothesized correlations between the OKI-DO instrument, a generic observation instrument [24] and psychosocial characteristics of the child (SDQ) [25]. We investigated multiple associations but decided against using, for example, Bonferroni correction. However, as

recommended by Nakagawa [42], we examined the effect sizes and found that almost all hypothesized correlations showed a small to medium effect size [31]. Furthermore, more than 90% of the hypothesized correlations were statistically significant or showed promising but not significant trend. The correlations between the SDQ questionnaire and the OKI-DO domains almost all showed non-significant trends. This could be due to the fact that the hypotheses were based on research with youth (11-18 years) with T1DM [6]. The OKI-DO domain "quality of instruction", however, showed some zero correlations with some of the generic domains, although we expected a (significant) correlation. The zero correlations between the OKI-DO domain "*quality of instruction*" and the generic domains "*quality of instruction*" and "*structure and limit setting*" may be explained by the different instructions that parents give when they are playing with their child compared to the instructions given during a medical procedure (like glucose monitoring). In this latter case, more assistance or a specific order in instructions may be required. This may have affected the results on this OKI-DO domain. Because the differences between instructions for play or during a medical procedure, we decided to keep the OKI-DO domain "*quality of instruction*".

Our sample size was lower than anticipated. Unfortunately, we were not able to include the 120 families we aimed to include [3]. Despite a participation rate of 64% (70% was expected), we included 77 families (this is approximately 10% of the total population of children with T1DM aged 0-7 years in the Netherlands [43]), although 15 hospitals participated in our study instead of 7 [3]. It is possible that families with problems during mealtime, glucose monitoring and/or insulin administration were reluctant to participate in our study. However, for reasons of confidentiality, we do not have non-response data and therefore are not able to further underpin this statement. Because the families in our sample did not encounter major problems during mealtime, glucose monitoring and insulin administration (low scores on '*negative behavior*', '*avoidance*', '*response to injection*', and high scores on '*limit setting*', '*respect for autonomy*', '*cooperative behavior*', see Table 2), we should regard our findings as preliminary evidence supporting the validity of the OKI-DO instrument. In future research, families who encounter problems during diabetes-specific situations should definitely be included to further examine the validity of the OKI-DO instrument.

Though widely used generic observation instruments [22-24] have been developed (based on theory or observations), studies that test the validity of observation instruments are scarce [28]. The preliminary validity of the OKI-DO instrument, however, was examined in the present study and showed positive indications for the preliminary validity. In the present study, we also investigated the inter-rater reliability of the OKI-DO instrument (weighted kappa was 0.73, indicating good inter-rater reliability). To further investigate the reliability of the

OKI-DO instrument, test-retest reliability could be examined in future research.

In our sample, 59.7% of the children had HbA_{1c} levels above the recommended ISPAD guideline of 58 mmol/mol or 7.5% [44]. This is in line with a recent large-scale European study [45] where 58% of the 27.035 participating children had a suboptimal HbA_{1c} level. Therefore we believe that the HbA_{1c} level is representative of other Western European children. However, most families that participated in our study were Caucasian (97%) and the majority of the children received pump therapy (82%). Therefore, we have to be cautious to generalize our findings to families with a different ethnic background and to children with multiple daily insulin injections. Furthermore, the educational level of the participants was generally higher than in the total Dutch population [46]. Approximately 32% of the adults in the Netherlands have an academic Bachelor's or Master's degree [46]. In this study 39% of the mothers and 100% of the fathers had an academic degree. Research shows that the educational level of parents is positively associated with the quality of parent-child interaction [47] and parenting strategies [48], so the findings of this study may be more applicable for parents with higher educational levels.

Conclusion

As diabetes-related family behaviors seem to be established in the early years post-diagnosis [9, 49], interventions should start as early as possible. The incidence of young children with T1DM is increasing [50] and early detection of problems and intervening in this young patient group is necessary. Observational research has shown that parents of children with T1DM have more parenting problems during mealtime [15, 17, 40] and there is a need for effective parenting strategies [14]. In future research, the OKI-DO observation instrument can be used to conduct studies that can help to determine whether parent-child interaction-patterns are associated with specific diabetes outcomes, such as glycosylated hemoglobin (HbA_{1c}) and psychosocial characteristics (such as quality of life). These results can than serve as a reference to determine, for example, whether interventions to improve parent-child interaction would be a meaningful intervention for families with a young child with T1DM. Furthermore, research showed that injection distress is more common in younger children and recently diagnosed children [51]. The OKI-DO instrument could enable scientists and clinical practitioners to evaluate interventions aimed at decreasing the injection distress for both parents and children and interventions aimed at optimizing the quality of parent-child interaction in families with a young child with T1DM.

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Chapter 5

The relationship between parenting stress and parent-child interaction with health outcomes in the youngest patients with type 1 diabetes (0-7 years)

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ABSTRACT

Aims
To test whether parenting stress and the quality of parent-child interaction were associated with glycemic control and QoL in young children (0-7 years) with T1DM.

Methods
77 families with a young child with T1DM were videotaped during mealtime (including glucose monitoring and insulin administration). Parent-child interactions were scored with a qualitative observation method specifically designed to assess parent-child interaction in diabetes-specific situations. Questionnaires assessed general and disease-related parenting stress, and (diabetes-specific (DS)) QoL. HbA_{1c} (glycemic control) was extracted from the medical records.

Results
Both general and disease-related parenting stress were associated with a lower (DS)QoL (r ranged from -0.39 to -0.70, $p<0.05$), but not with HbA_{1c} levels. Furthermore, with regard to the parent-child interaction, emotional involvement of parents ($r=0.23$, $p<0.05$) and expressed discomfort of the child ($r=0.23$, $p<0.05$) during mealtime (including glucose monitoring and/or insulin administration) were related to suboptimal HbA_{1c} levels. There was no clear pattern in the correlations between parent-child interaction and (DS)QoL.

Conclusions
The results of the present paper support the notion that diabetes does not only affect the child with T1DM: T1DM is a family disease, as parenting factors (like stress and parent-child interactions) are associated with important child outcomes. Therefore, it is important for health care providers to not only focus on the child with T1DM, but also on the family system.

Introduction

Nowadays, the number of children being diagnosed with T1DM is growing, with an overall annual increase of almost 4% [1]. This number is particularly growing in the youngest age group [1-3]. In children with T1DM, achieving an optimal HbA_{1c} level (≤ 58 mmol/mol or 7.5% [4]) is an important treatment goal. Adequate glycemic control helps to avoid or delay the onset of long-term micro- and macro vascular complications, such as neuropathy, retinopathy, nephropathy and cardiovascular diseases [5]. In addition to these long-term consequences, suboptimal glycemic control is also associated with short-term consequences like a negative effect on school performances [6, 7] and child behavior problems [8, 9]. Despite all efforts, more than half of the children with T1DM still do not reach this optimal HbA_{1c} level [10, 11]. Having T1DM can also have an adverse effect on children's quality of life (QoL) [12], therefore, health care providers not only focus on reaching an optimal HbA_{1c} level, but also on maintaining or enhancing a good QoL in children with T1DM [13]. The literature on QoL in children with T1DM is contradictory as some studies report an obvious impairment in QoL in children with T1DM compared to healthy peers (e.g. [12, 14]), whereas others report a similar level of QoL compared to healthy peers or that children with T1DM even show adaptive outcomes (e.g. [15, 16]). A recent systematic review concluded that the differences in QoL between children with T1DM and healthy peers were, on average, only minimal; although diabetes-specific (DS) QoL problems (e.g. worries, impact on daily functioning) were certainly present [17].

When young children are diagnosed with T1DM, parents have complete responsibility for the daily diabetes-management (assessing blood glucose levels, administering insulin, regulating food intake, and guarding these parameters in conjunction with the level of physical activity) of their child [18, 19]. Because of these many proceedings, having to think constantly about the correct amount of insulin/carbohydrates, and adjustments during the day without help from daycare or school, parents might consider this as having an 'extra job'. Therefore, it is not surprising that many parents of young children with T1DM report increased levels of parenting stress [20] and difficulties in parent-child interactions [21]. These parenting factors (parenting stress and parent-child interaction) are strongly linked to parent-, child- and contextual characteristics [22, 23], like the Process Model of Belsky states [24]. As parent-, child-, and contextual factors are linked to diabetes outcomes [20, 25, 26], parenting stress and parent-child interaction may also be related to the HbA_{1c} level and QoL of the child.

A recent review [27] has described that higher levels of parenting stress were associated with suboptimal HbA_{1c} levels in school-aged children and adolescents (age range 7-17 years).From the same review it appeared that in

younger children (aged 0-11 years), parenting stress was not or even negatively associated with HbA_{1c} levels, indicating that higher levels of stress were related to more optimal HbA_{1c} levels [27]. This discrepancy in findings between parenting stress and HbA_{1c} levels in parents of school-aged children and adolescents versus parents of young children could be due to the fact that parents of older children have a shared responsibility, while parents of younger children have a full responsibility for the care of their children with T1DM. Increased levels of parenting stress could indicate higher levels of involvement in the diabetes regimen and therefore more optimal HbA_{1c} levels in young children [27]. The level of parenting stress has not been associated with child QoL often, as the same review [27] only found one study that associated higher levels of parenting stress with a lower child QoL. This study, however, included only children between 12-17 years of age [28]. It is important to gain more knowledge about the associations between parenting factors and child outcomes, as parenting stress might be beneficial for HbA_{1c} levels in young children, but not for child QoL [27]. Therefore, knowledge about the association between parenting stress and child QoL in young children with T1DM is insufficient.

Only a few studies have investigated the quality of parent-child interaction in children with T1DM, showing that particularly parental over-involvement, parental restrictiveness, parental hostility, conflicts and negative communication are significantly associated with suboptimal HbA_{1c} levels (e.g. [18, 29-34]) and a lower reported QoL (e.g. [35, 36]). In contrast, positive maternal communication, positive reinforcement, emotional support, parental warmth and caring behavior appeared to be significantly associated with more optimal HbA_{1c} levels (e.g. [32, 33, 37]) and a better reported QoL (e.g. [32, 38]) of children and adolescents with T1DM. However, the aforementioned studies mainly focused on older children (aged 8-20 years) [29, 30, 32-38] or used rather diverse age groups (aged 4-14) [18, 31] and therefore, research in exclusively (very) young children is limited. We have only found one study that directly observed parent-child interaction in younger children (aged 2-8 years) [39]. This study showed that ineffective parenting during mealtime (i.e., coaxes, interrupted commands and physical prompts) was significantly related to suboptimal HbA_{1c} levels [39]. Currently, research on the parent-child interaction in younger children including also QoL as an outcome factor is lacking.

Research examining the associations among parenting stress, parent-child interaction and child outcomes in young children with T1DM is scarce and urgently needed. More insight in this area may contribute to the development of new, effective interventions. Therefore, the present study was conducted to test whether, and how, parenting stress and the quality of parent-child interaction were related to the HbA_{1c} level and QoL in young children (0-7 years) with T1DM.

METHODS

Patients and procedure

Children (aged 0-7 years) diagnosed with T1DM more than 6 months and their parents were recruited from 15 hospitals/institutions in the middle and southern part of the Netherlands (including Kidz&Ko, a partnership between seven pediatric diabetes clinics, and Diabeter, a national center for pediatric and adolescent diabetes care and research). Parents who lacked basic proficiency in Dutch, and children with an Autism Spectrum Disorder, Down syndrome and/or other mental disabilities (n=17) were excluded. Of the 121 eligible families, 77 families (64%) agreed to participate. After receiving written informed consent, the mealtime situation (including glucose monitoring and insulin administration) was videotaped during a home visit (for a detailed description of the procedure of the home-visits see Nieuwesteeg et al. 2014 [40]). Furthermore, both parents were asked to complete a questionnaire assessing (socio)demographic and clinical variables, general and disease-specific parenting stress and generic and diabetes-specific (DS) child QoL. In the present study, only scores of the questionnaires of the parent who was videotaped during the interaction were used. Of the 77 observed parents (96% mothers), 70 parents (96% mothers) completed the questionnaire about general parenting stress and DSQoL of their child, 68 parents (96% mothers) completed the questionnaire about the generic QoL of their child, and 64 parents (94% mothers) completed the questionnaire about disease-related parenting stress. The study was approved by the Medical Ethical Review board of St. Elisabeth Hospital Tilburg (date: 25-05-2010).

Measures

<u>(Socio)demographic and clinical variables</u>

The first part of the questionnaire included items involving (socio) demographic characteristics (i.e., gender and age of the child, marital status and educational levels of both parents) and clinical characteristics (i.e., treatment regimen, frequency of blood glucose monitoring (average number of assessments/day), and years since diagnosis).

<u>Metabolic control</u>

Glycemic control (HbA_{1c} level, measured closest to the home visit) was determined at the hospital where the child was treated for T1DM and extracted from the medical records of the children, after receiving written consent from the parents.

Parent-child interaction

To assess the quality of parent-child interaction, the mealtime situation (including glucose monitoring and insulin administration) was videotaped in all participating families during a home visit. The videotapes were scored by an observer (AN) with the qualitative OKI-DO observation instrument, which is specifically developed for children with T1DM, to assess the quality of parent-child interaction during diabetes-specific situations [40]. This observation instrument comprises ten domains, including four parent domains ("emotional involvement", "limit setting", "respect for autonomy", and "quality of instruction"), four child domains ("negative behavior", "avoidance", "cooperative behavior", and "child's response to injection"), and two family-domains ("emphasis on diabetes", and "mealtime structure"). The parent-child dyad will receive a score (1-5) on each domain, in which higher scores reflect more of the behavior (e.g., a high score on 'emotional involvement' means the parent is highly emotional involved, and a high score on 'negative behavior' means the child shows a lot of negative behavior). An example of the rating scale "respect for autonomy" can be rated varying from score 1: '*The caregiver receives this score if he/she fully determines what should happen without explaining anything to the child and with a visible lack of respect for the autonomy. For example: the caregiver just takes the finger of the child to check the glucose, (harshly) 'pulls' the child in the correct position to operate the insulin pump or determines (without consulting or warning the child) where and when the insulin injection takes place, the caregiver fully determines what and how much the child eats. If the child is (rather) independent in managing his/her diabetes, the caregiver receives this score if he/she repeatedly interferes when the child is managing his/her diabetes, while it is clear from the observation that the child can perform everything on its own*', to score 5: '*The caregiver receives this score if he/she praises initiatives of the child and encourages the child to make decisions on his own regarding his/her diabetes. The child may, for example, read the glucose meter, operate the insulin pump or determine where and when the insulin injection takes place (the caregiver could of course check the things his/her child does, but is herein not at all intrusive). Everything is determined in consultation with the child and the child is treated with respect.*' To assess the inter-rater reliability of the observations, a second observer (HvB) scored 20% of the videotapes independently of the first observer. The agreement with the first observer was high as weighted kappa was 0.73, indicating a good inter-rater reliability. Research shows encouraging indications for the usability, reliability and preliminary validity of the OKI-DO instrument to assess parent-child interaction in young children with T1DM during mealtime (including glucose monitoring and insulin administration) [40].

General parenting stress

General parenting stress was assessed with the short Dutch version of the Parenting Stress Index [41, 42]. This is a 17-item self-report measure, in which parents report how much they agree on the propositions about stress in the parent-child system. The items are rated from 1 (strongly disagree) to score 4 (strongly agree). All items were summed to get a total score (α =0.92). Higher scores reflect more general parenting stress. The 17-item version has shown good reliability [43].

Disease-related parenting stress

Disease-related parenting stress was assessed with a validated, 42-item self-report measure, the Pediatric Inventory for Parents (PIP) [44, 45]. Parents are asked to describe both the frequency and difficulty of experienced disease-related parenting stress across four domains: "communication" (for example: with the medical team, partner, child), "emotional distress" (for example: quality of sleep, effect on mood), "medical care" (for example: treatment demands) and "role functioning" (for example: being able to go to work). All items are scored on a 5-point Likert scale on both *frequency* and *difficulty*. The scale scores were summed to get a total *frequency* (α = 0.93) and total *difficulty* score (α = 0.93). Higher scores reflect more frequency and difficulty in disease-related parenting stress. Adequate internal consistency and construct validity of the original version of the PIP have been reported [44].

Quality of life of the children

Generic QoL was measured with the TNO-AZL Preschool Quality Of Life questionnaire or TAPQOL [46] in children in the age of 1 through 5 years of age and the TNO-AZL Child Quality Of Life questionnaire or TACQOL [47] in children of 6 and 7 years of age. These multidimensional questionnaires are proxy measures, as they assess the parent's perceptions of health-related QoL in (preschool) children. The TAPQOL includes 43 items constituting 12 scales covering aspects of QoL: seven health-related scales: stomach problems, skin problems, lung problems, sleeping problems, appetite, motor functioning, and communication (these seven scales were summed and then averaged (health-related QoL), α = 0.80), and five psychosocial-related scales: liveliness, positive mood, problem behavior, anxiety, and social functioning (these five scales were summed and then averaged (psychosocial QoL), α = 0.75). The TACQOL includes 63 items constituting seven scales covering aspects of QoL: five health-related scales; pain and symptoms, motor function, autonomy, cognitive functioning, interaction with parents and peers (these five scales were summed and then averaged (health-related QoL), α = 0.94), and two psychosocial-related scales; experience of

positive and negative emotions (the two scales were summed and then averaged (psychosocial QoL), $\alpha = 0.84$). In both questionnaires, the parent indicates to what extent specific problems of the health-related functioning scales occurred in the past few weeks, with three response categories: 'never', 'sometimes', and 'often'. If a problem occurs, the parents are asked how the child is feeling: '(very) good', 'not so good', 'pretty bad', and 'bad'. For each item, the two answers are combined into a single item score ranging from 0 to 4 ('never' 4 and 'sometimes' or 'often' combined with '(very) good' 3, 'not so good' 2, 'pretty bad' 1, and 'bad' 0). With the psychosocial-related scales, the parents indicate on a Likert scale whether a certain emotion in their child has appeared in the last few weeks ('never', 'sometimes', 'often'). Item scores for the psychosocial-related scales run from 0 to 2. All TAPQOL and TACQOL scales are linearly transformed to 0-100 scales; higher scores will correspond to a better QoL. Research showed that the TAPQOL and the TACQOL are reliable and valid questionnaires [48, 49].

Diabetes-specific QoL (DSQoL) was measured with a child self-report questionnaire (Smiley Faces) for young children, composed by the Hvidøre Study Group [50]. This questionnaire has been modified in a proxy-report form with permission of the authors, so that the parents can complete the questionnaire for their child. The questionnaire comprises 19 items about feelings of the child in relation with his or her diabetes (for example, items about administering insulin, health, leisure time, and school/nursery/daycare). The items are rated on a 5-point Likert scale. Scores are (re)coded so that a higher score corresponds to a better QoL. All items were summed to get a total diabetes-specific QoL score ($\alpha = 0.86$). The originally child self-report questionnaire showed good reliability and validity [50].

Statistical analyses

Statistical analyses were conducted using the Statistical Package for the Social Sciences (SPSS, version 19). Frequencies and descriptive statistics were used to present the (socio)demographic and diabetes-related characteristics of the participating families. Pearson's correlation coefficients were calculated to examine the strength of the relationships between parenting stress and the parent-child interaction with HbA_{1c} levels and (DS)QoL. Because different QoL questionnaires were used for children aged 0-5 and 6-7, we have examined the Pearson's correlations for these questionnaires (generic and DS QoL) separately. The results of the correlations were considered statistically significant when $p \leq 0.05$. Furthermore, the effect sizes were examined. According to Cohen, r of 0.10, 0.30, and 0.50 are considered as small, medium and large effects, respectively [51].

RESULTS

Sociodemographic and clinical data

Table 1 summarizes the characteristics of participating parents and children. Among the participating children, there were 41 boys (53%). The children with T1DM had a mean age of 5 years (SD = 1.5, range: 2-7 years). Most children (82%) received pump therapy. On average, parents monitored their child's blood glucose 6 times a day (range: 2-20). The mean HbA_{1c} level of the children, measured closest to the home visit, was 59 mmol/mol or 7.6% (range 32-80 mmol/mol or 5.1%-9.5%).

Most of the 70 parents (96% mothers) who completed the questionnaires, were cohabiting or married/registered partners (7% of the mothers and 0% of the fathers were single). More than half of the participating mothers (54%) had a higher educational level (i.e., approximately 12 years of formal education), while all fathers (100%) had a Bachelor's or Master's degree (i.e., approximately 15 years of formal education).

Table 1: (socio)Demographic and clinical characteristics of young children with type 1 diabetes and their parents.

			N (%)	M	SD
Children	Sex	Boys	41 (53%)		
		Girls	36 (47%)		
	Age (years)	(range 2-7)		5.12	1.52
	HbA_{1c}	(range 32mmol/mol-80mmol/mol)	77	59	9.03
		(range 5.1% - 9.5%)	77	7.6%	0.8%
	Treatment	Insulin pump	63 (82%)		
		Multiple daily insulin injections	14 (18%)		
	Blood glucose monitoring	Times a day (range 2-20)		6.08	2.69
	Years since diagnose	(range 1-6 years)		2.61	1.44
Parents	Total	Mothers observed	74 (96%)		
		Fathers observed	3 (4%)		
	Marital status (mothers)	Single	5 (7%)		
		Cohabiting	10 (13%)		
		Married / registered partners	52 (70%)		
		Missing	7 (10%)		
	Marital status (fathers)	Single	0 (0%)		
		Cohabiting	1 (33%)		
		Married / registered partners	2 (67%)		
		Missing	0 (0%)		
	Educational level (mothers)	Primary education	1 (1%)		
		About 12 years of formal education	36 (49%)		
		15-16 years of formal education	29 (39%)		
		Other	1 (1%)		
		Missing	7 (10%)		
	Educational level (fathers)	Primary education	0 (0%)		
		About 12 years of formal education	0 (0%)		
		15-16 years of formal education	3 (100%)		
		Other	0 (0 %)		
		Missing	0 (0%)		

Associations between parenting stress and the quality of parent-child interaction with HbA_{1c} *levels and (diabetes-specific) quality of life.*

Table 2 summarizes the Pearson correlations between general parenting stress, disease-related parenting stress and the domains of the quality of parent-child interaction with the HbA_{1c} level and (DS)QoL of the children.

Results showed that general and disease-related parenting stress were significantly negatively associated with both generic QoL (correlations ranging from -0.40 to -0.56, medium to large effect sizes) and diabetes-specific QoL (correlations ranging from -0.39 to -0.70, medium to large effect sizes). HbA_{1c} level did not correlate significantly with general or disease-related parenting stress. These results indicate that elevated levels of both general and disease-related parenting stress were associated with lower (DS)QoL of young children with T1DM.

Furthermore, only the OKI-DO domains *emotional involvement* (r = 0.23, small effect size) and *child's response to injection* (r = 0.23, small effect size) were significantly positively associated with the HbA_{1c} level of the child, which indicates that parents with children with higher HbA_{1c} levels were more emotionally involved and that children with higher HbA_{1c} levels expressed more discomfort during glucose monitoring (for example more tension in body, tightening eyes, crying or resisting behavior). A better *quality of instruction* was associated with a better generic QoL in children aged 6-7 (r = 0.41, medium effect size), and more *emphasis on diabetes* during the meal was significantly correlated with a better DSQoL in children aged 0-5 (r = 0.36, medium effect size).

Table 2: Relationships between (disease-related) parenting stress, the OKI-DO domains and child (health) outcomes.

		0-5 years			6-7 years		
	HbA_{1c}	**Generic QoL**		**DSQoL**	**Generic QoL**		**DSQoL**
		Health related	**Psycho-social**	**Total**	**Health related**	**Psycho-social**	**Total**
	r	**r**	**r**	**r**	**r**	**r**	**r**
Parenting stress							
General total	0.12	-0.13	-0.18	**-0.42 ****	**-0.53 ****	**-0.56 ****	**-0.70 ****
Disease-related frequency total	-0.15	-0.19	-0.18	**-0.39 ***	**-0.56 ****	**-0.40 ***	**-0.69 ****
Disease-related difficulty total	-0.09	-0.16	-0.10	-0.33	**-0.52 ***	-0.27	**-0.70 ****
Parent-child interaction							
Emotional involvement	**0.23***	-0.12	0.13	-0.04	0.02	-0.01	0.25
Limit setting	0.08	-0.17	0.07	0.03	-0.13	-0.12	0.02
Respect for autonomy	-0.05	-0.17	-0.11	-0.22	0.08	0.19	0.23
Quality of instruction	-0.12	-0.16	-0.08	0.02	0.13	**0.41 ***	0.19
Negative behavior	0.03	0.20	0.08	0.26	0.35	0.22	0.15
Avoidance	-0.02	0.15	0.16	0.08	-0.06	-0.30	-0.16
Cooperative behavior	0.11	-0.12	0.03	-0.02	0.18	-0.05	0.16
Child's response to injection	**0.23***	-0.16	-0.01	-0.14	0.03	-0.32	-0.13
Emphasis on diabetes	0.14	0.07	0.22	**0.36 ***	0.24	0.08	-0.10
Mealtime structure	-0.02	-0.19	-0.10	-0.11	-0.25	-0.19	-0.07

* p<.05
** p<.01

DISCUSSION

For health care providers, maintaining or enhancing a good QoL of children with T1DM is as important as guarding or achieving good glycemic control [13]. However, research examining factors that are associated with HbA_{1c} levels and (DS)QoL in young children with T1DM is still scarce. The present study focused on this youngest patient group and found that higher levels of (disease-related) parenting stress were associated with a lower (DS)QoL of the child and that emotional involvement and the child's response to injection were associated with lower HbA_{1c} levels.

The results showed that parenting stress was not associated with HbA_{1c} levels. A previous study, however, concluded that higher levels of disease-related parenting stress were associated with more optimal HbA_{1c} levels in young children with T1DM [52]. An explanation of the discrepancy in findings between our study (no significant correlation) and the study of Stallwood [52] might be explained by type of insulin treatment: parents generally experience less disease-related parenting stress when their child is on insulin pump therapy [53, 54]. In the study of Stallwood [52] none of the children used pump therapy, in contrast to 84% of the children in the present study. Future research should examine the association between glycemic control and disease-related parenting stress in a sample with a comparable amount of children using multiple daily insulin injections versus insulin pump therapy to see whether there is or is not a significant association between disease-related parenting stress and HbA_{1c} levels. If there indeed is a significant association, appropriate interventions in order to lower disease-related

parenting stress could be part of the treatment in lowering HbA_{1c} levels.

Furthermore, parents who experienced higher levels of (disease-related) parenting stress reported lower child QoL, especially DSQoL (e.g., problems with insulin administration, glucose monitoring, energy level, health etc.). This is the first study that has examined this relationship in young children with T1DM. Because of these results, health care providers should not only be aware of the (DS)QoL of the child, but also the perceived (disease-related) parenting stress of the parents as higher levels of (disease-related) parenting stress are associated with lower (DS)QoL. Therefore, health care providers should monitor the level of (disease-related) parenting stress regularly in order to avoid a low QoL of the child.

Parents were more emotionally involved when their child had a suboptimal HbA_{1c} level. Research with older children and adolescents with T1DM also showed that a suboptimal HbA_{1c} level was positively associated with emotional involvement of the parents [29] and involvement in diabetes care [30]. However, the causal direction of this association remains unclear. It could be that parents are more emotionally involved when trying to get a more optimal HbA_{1c} level and thereby delaying the onset of long- and short-term complications [29]. Otherwise, it could also be true that parents who are highly emotionally involved during diabetes-specific situations deliberately set higher HbA_{1c} targets for their child, because of fear of hypoglycemia [55].

Furthermore, the results showed that children with a high HbA_{1c} level expressed more discomfort during glucose monitoring and/or insulin administration (for example more tension in body, tightening eyes, crying or resisting). Displaying more discomfort could be due to some form of needle-phobia or a low pain threshold. When children experience needle-phobia or have a low pain threshold, parents might postpone or omit insulin injections or glucose monitoring in order to avoid these stressful and unpleasant situations, which could result in suboptimal HbA_{1c} levels [56]. It could also be true that a high HbA_{1c} level might 'worsen' the diabetes for both parent and child, and that the child, due to an emphasize on their disease, experiences more discomfort during glucose monitoring and/or insulin administration.

Remarkably, the other domains of parent-child interaction around the diabetes situation were not related to HbA_{1c} level. Based on results of a previous study with young children [39] and studies with older children [18, 29-34], showing that ineffective parenting strategies and child misbehavior were significantly correlated with less optimal HbA_{1c} levels, we expected that, for example, less respect for autonomy, or more negative behavior of the child would be associated with suboptimal HbA_{1c} levels. This discrepancy in findings might be due to different sample characteristics, as our sample had more optimal

HbA_{1c} levels (7.6% or about 59 mmol/mol in our study versus >8% or >64 mmol/mol [18, 29, 30, 34, 39]). Furthermore, the children in our sample were much younger than in most other studies [18, 29-34]. Also, it is possible that families who encountered problems (like child misbehavior) during the mealtime situation (including glucose monitoring and insulin administration) were reluctant to participate in the present study. Our sample consisted of rather high-functioning families, meaning that they did not encounter major problems during mealtime, glucose monitoring and insulin administration [40].

The quality of parent-child interaction did not show clear patterns of correlations with (DS)QoL. Previous research with youth with T1DM showed that parent-child behaviors (e.g., diabetes-specific family conflict, warmth) were related to QoL [36, 38]. The parent-child behaviors in these studies were collected through self-report questionnaires [36, 38]. Weisberg-Benchell [35] also examined the relationship between QoL and parent-child behavior, but they used both self-report questionnaires and observations to assess parent-child behaviors. While the self-reported parent-child behaviors did correlate with QoL, the observed parent-child behaviors did not [35]. This highlights the importance of using direct observations in examining the quality of parent-child interaction in future studies, with an observational instrument like the OKI-DO instrument, as self-reports might reflect a more subjective view.

The present study has some limitations that need to be described. Research has shown that diabetes centers not only differ in structural issues, but also at an educational level and guidance regarding diabetes, which could influence glycemic control [57]. Because we included children from 15 different hospitals/institutions, the diabetes education and guidance these parents and children receive(d) could be very different. Furthermore, our sample consisted of almost only Caucasian participants (97%), participation rate among fathers was very low, most of the parents had a relatively high educational level, which is not a fair representation of the population in the Netherlands [58]. As the educational level and socioeconomic status of parents is positively associated with the quality of parent-child interaction [59] and parenting strategies [60, 61], the results of the present study may not be generalized to parents with a lower educational level. Also, because of the videotaped home visits, it is possible that families who frequently experience problems during mealtime, glucose monitoring and/or insulin administration were reluctant to participate in our study, which might have led to biased results. Future research, therefore, should examine whether a causal relationship exists between the quality of the parent-child interaction and (disease-related) parenting stress with (health) outcomes of children with T1DM, including families of different ethnic backgrounds, parents with lower educational levels, and families who encounter problems during diabetes-specific situations.

A strength of the present study is the use of the OKI-DO instrument, as observations of "real life" parent-child interactions and scored by an independent observer can provide more objective data to assess the quality of parent-child interaction than using self-report questionnaires or interviews [40]. The OKI-DO instrument could be used by health care providers to evaluate interventions (like video interaction guidance) aimed at optimizing the quality of parent-child interaction and/or lowering the parenting stress in order the influence child outcomes. Furthermore, as recommended by Nakagawa [62], we refrained from correcting for examining multiple associations to be able to give an overview of all the associations that were examined as we were interested in all possible associations between the included variables. Another strength is the support from several large diabetes clinics in the Netherlands. Therefore, it was possible to focus on younger children with T1DM and their families, which makes this study innovative as this patient group is understudied.

The results of the present paper support the notion that diabetes does not only affect the child with T1DM: T1DM is a family disease, as parenting factors (like stress and parent-child interactions) are associated with important child outcomes. Therefore, it is important for health care providers to not only focus on the child with T1DM, but also on the family system.

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Chapter 6

Pediatric parenting stress in fathers and mothers of young children with type 1 diabetes: a longitudinal study

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ABSTRACT

Objective
To compare levels of pediatric parenting stress between fathers and mothers of young children with type 1 diabetes (T1DM) and study the course of pediatric parenting stress over time.

Methods
112 parents (56 mothers and 56 fathers) of young children with T1DM (0-7 years) completed the Pediatric Inventory for Parents assessing pediatric parenting stress (frequency and difficulty scores on the subscales: communication, emotional distress, medical care, role functioning and total score), of whom 44 mothers (79%) and 31 fathers (55%) completed the questionnaire again one year later. Independent and paired sample t-tests were used to examine the differences between fathers and mothers and change over time, respectively. Furthermore, Cohen's d effect sizes were calculated.

Results
At the first measurement occasion, mothers scored significantly higher than fathers on the pediatric parenting stress subscales: communication frequency and difficulty, emotional distress frequency and difficulty, medical care frequency, and total score frequency and difficulty (d ranged from -0.44 to -0.56). Furthermore, it appeared that fathers reported to experience a decrease in medical care frequency (d=0.10) and an increase in emotional distress difficulty (d=-0.32) and total score difficulty (d=-0.29), whereas mothers reported to experience a decrease of pediatric parenting stress in emotional distress frequency, medical care frequency and total score frequency (d ranged from 0.31-0.66) over a one year period.

Conclusions
These results show that within families with a young child with T1DM, burden of care increases in fathers and decreases in mothers, suggesting an increased care responsibility of the fathers when their child with T1DM grows up.

Background

Parenting children can be stressful, but parenting a child with a chronic illness, like asthma, cancer, cystic fibrosis, diabetes, epilepsy, juvenile rheumatoid arthritis, and/or sickle cell disease, is perceived as even more stressful as examined by a meta-analysis by Cousino and Hazen [1]. On top of general parenting stress, parents of children with a chronic illness worry about their child's health, treatment demands, and communication about the illness with their child and the medical team, which is defined as pediatric parenting stress [2]. Although all parents of children with a chronic illness are burdened with certain concerns and treatment demands, Hullmann et al. [3] found that parents of children with type 1 diabetes mellitus (T1DM) (M = 75.70, SD = 1.70) reported significantly higher levels of parenting stress than parents of children with cancer (M = 69.22, SD = 1.96) or cystic fibrosis (M = 71.69, SD = 2.67). According to Hullmann et al. [3] this could be explained by the complex treatment regimen of T1DM. This regimen consists of monitoring the blood glucose level several times a day, administering insulin, regulating food intake, and guarding these parameters in conjunction with the level of physical activity of their child. Because parents of young children with T1DM bear the full responsibility for their child's treatment demands, they often experience higher levels of stress than parents of older children with T1DM [4-6].

A recently published review provides a comprehensive overview of the current studies regarding the level of pediatric parenting stress that is experienced by parents of children with T1DM [1]. However, the studies that were included in that review mainly focused on parents with older children (7-19 years) [5, 7-9], or used a wide age range (1-17 years) [4, 10]. Moreover, the few studies that focused on parents of younger children (0-9 years), mainly reported on mothers' experienced pediatric parenting stress [4, 6, 11-14]. Fathers of young children with T1DM are generally being overlooked when pediatric parenting stress is examined. In the review by Cousino and Hazen [1] only one study is included that studied emotional distress in fathers of young children (aged 2-6) with T1DM. Results of that study showed that fathers experienced relatively mild levels of pediatric parenting stress [15]. Unfortunately, in that study information about the mothers was lacking [15]. Therefore, research on pediatric parenting stress of both fathers and mothers of young children with T1DM is lacking.

Based on earlier research in parents of older children with T1DM (7-14 years) [8, 10], it can be expected that mothers experience higher levels of pediatric parenting stress than fathers. Mothers are usually responsible for most of the diabetes-management of their child [16] and, therefore, might experience higher levels of pediatric parenting stress than fathers. Currently, there seems to be a lack of information about the differences in pediatric parenting stress in

mothers and fathers of young children with T1DM.

Even though literature has convincingly shown that parents of young children with T1DM have elevated levels of pediatric parenting stress [4-6], information about pediatric parenting stress in these families over time is scarce. To our knowledge, one study has examined the course of pediatric parenting stress in parents of young children (4-7 years) with T1DM over time and found that parents reported less pediatric parenting stress after transition from multiple daily insulin injections to continuous subcutaneous insulin infusion [4]. This study, however, included only a small number of parents (n=24), and has examined the change in pediatric parenting stress after their child has changed treatment regimen.

The present study compares levels of perceived pediatric parenting stress between fathers and mothers of young children with T1DM, and also examines the course of pediatric parenting stress in these fathers and mothers over a one year period. Our hypotheses, based on research with parents with older children with T1DM [8, 10], is that mothers report higher levels of pediatric parenting stress than fathers. Furthermore, based on a study with parents of young children with T1DM who changed treatment regimen [4], we expect that parents experience less pediatric parenting stress over time as they get more used to the daily intensive treatment demands.

METHODS

Patients and procedure

Mothers and fathers of young children (0-7 years) with a diagnose T1DM for > 6 months were recruited from 15 hospitals/institutions in the middle and southern part of the Netherlands (including Diabeter, a national center for pediatric and adolescent diabetes care and research, and Kidz&Ko, a partnership between seven pediatric diabetes clinics). Parents who lacked basic proficiency in Dutch and parents of children with a mental disability, Down syndrome or an Autism Spectrum Disorder were excluded (n = 17). During a home visit (see Nieuwesteeg et al. [17]) data regarding (socio)demographic and clinical variables, and also pediatric parenting stress was collected from both the father and the mother. Two families did not want to participate with the home visit, but did agree to complete the questionnaire, resulting in a total of 62 fathers and 72 mothers who completed (some part of) the questionnaire. Because we wanted to know whether the perceived pediatric parenting stress would change after one year in these fathers and mothers, we sent all parents who had participated in the first measurement wave (both the father and mother) the same questionnaire by mail one year after the home visit and asked them to return the questionnaire within two weeks. When the questionnaires were not returned within three

weeks, the families were contacted by phone and were asked to send the questionnaires or to withdraw from the study if they did not want to participate anymore. In total, 31 fathers (50%) and 44 mothers (61%) completed the questionnaire at the second measurement wave one year later. The study was approved by the Medical Ethical Review board of St. Elisabeth Hospital Tilburg (date: 25-05-2010) and in conjunction with the Helsinki Declaration on human research.

Measures

(Socio)demographic and clinical variables

Both the father and the mother of the child completed a questionnaire with (socio)demographic characteristics, including gender and age of the child, relationship with the child (father or mother), marital status (single, cohabiting, or married/registered partners) and educational levels of both parents (primary education, about 12 years of formal education, 15-16 years of formal education, or other) and clinical characteristics, including treatment regimen (insulin pump or multiple daily insulin injections), frequency of blood glucose monitoring a day, and HbA_{1c} level.

Pediatric parenting stress

Pediatric parenting stress was assessed with the 42-item Pediatric Inventory for Parents (PIP) [2, 18]. Parents are asked to describe on a 5-point Likert scale both the frequency and difficulty of pediatric parenting stress across four subscales: "Communication" (for example: with the medical team, partner, child, score range: 9-45), "Emotional distress" (for example: quality of sleep, effect on mood, score range: 15-75), "Medical care" (for example: treatment demand, score range: 8-40) and "Role functioning" (for example: being able to go to work, score range: 10-50). The scale scores were summed to get a total *frequency* and total *difficulty* pediatric parenting stress score (score range: 42-210). Higher scores reflect higher frequency and increased difficulties in terms of pediatric parenting stress. Adequate internal consistency and construct validity of the original version of the PIP have been reported [2]. In the present study, α's for all *frequency* and *difficulty* subscales for fathers and mothers on both time points were ≥ 0.70 (except for the Medical Care frequency subscale (α ranges between 0.63 and 0.68 for fathers and mothers on both time points, which is in line with previous research validating the PIP in mothers of children with T1DM as they also found an $\alpha < 0.70$ in the Medical care frequency scale [9]), and α's for the total *frequency* and *difficulty* scores were ≥ 0.90 for mothers and fathers on both time points. Furthermore, all subscales of the PIP on both measurement occasions in both fathers and mothers correlated significantly with each other.

Statistical analyses

The Statistical Package for the Social Sciences (SPSS, version 19.0) was used to conduct the statistical analyses. To compare the mean scores of fathers and mothers at the first measurement occasion, paired sample t-tests were used. When comparing the mean scores of fathers and mothers over time, paired sample t-tests were used. Furthermore, Cohen's *d* effect sizes were calculated to examine the standardized differences between the means. According to Cohen, effect sizes *d* of 0.20, 0.50, and 0.80 can be considered small, medium, and large, respectively [19].

Results

Sociodemographic and clinical data

In total, 72 mothers (54%) and 62 fathers (46%) completed the questionnaire at the first measurement occasion, representing 72 separate families. Because of missing items, we could only include 56 fathers (90%) and their spouses (78%).Table 1 summarizes the characteristics of these participating parents (56 mothers and 56 fathers) and their children (n = 56). Most of the mothers and fathers were cohabiting or married/registered partners (only 4% of the mothers and fathers were single). More than half of the participating mothers (57%) and fathers (52%) had a higher educational level (i.e., approximately 12 years of formal education), while 41% of the mothers and 48% of the fathers had a Bachelor's or Master's degree (i.e., approximately 15 years of formal education). Their children had a mean age of 5 years (SD= 1.6, range: 2-7 years) 52% were boys. Most children (82%) received pump therapy and had the diagnose T1DM for more than 2 years. On average, parents monitored their child's blood glucose 6 times a day (range: 2-20). The mean HbA_{1c} level of the children was 59 mmol/mol or 7.6% (range 32-80 mmol/mol or 5.1%-9.5%).

Table 1: Sociodemographic and clinical characteristics of young children with type 1 diabetes and their parents at the first measurement occasion.

			N (%)	M	SD
Children	Sex	Boys	29 (52%)		
		Girls	27 (48%)		
	Age (years)	(range 2 - 7)		5.18	1.61
	Years since diagnose	(range > 6 months - 7 years)		2.66	1.48
	HbA1c	(range 32 mmol/mol - 80 mmol/mol)		58.8	9.65
		(range 5,1% - 9,5%)		7.6	0.88
	Treatment	Insulin pump	46 (82%)		
		Multiple daily insulin injections	10 (18%)		
	Blood glucose monitoring	Times a day (range 2-20)		6.27	2.61
Parents	Total	Mothers	56 (50%)		
		Fathers	56 (50%)		
	Marital status (mothers)	Single	2 (4%)		
		Cohabiting	8 (14%)		
		Married / registered partners	46 (82%)		
	Marital status (fathers)	Single	2 (4%)		
		Cohabiting	7 (12%)		
		Married / registered partners	47 (84%)		
	Educational level (mothers)	Primary education only	0 (0%)		
		About 12 years of formal education	32 (57%)		
		15-16 years of formal education	23 (41%)		
		Other	1 (2%)		
	Educational level (fathers)	Primary education only	0 (%)		
		About 12 years of formal education	29 (52%)		
		15-16 years of formal education	27 (48%)		
		Other	0 (0%)		

Differences between fathers and mothers in pediatric parenting stress at first measurement

Table 2 summarizes mean scores and standard deviations on the subscales and total scales assessing pediatric parenting stress of fathers and mothers at the first measurement occasion. When comparing the mean scores of fathers and mothers, a significant difference was found on the scales Communication *frequency* ($t_{(54)} = -3.605$, $p = 0.001$, $d = -0.50$), Communication *difficulty* ($t_{(51)} = -3.997$, $p < 0.001$, $d = -0.51$), Emotional distress *frequency* ($t_{(54)} = -3.994$, $p < 0.001$, $d = -0.51$), Emotional distress *difficulty* ($t_{(51)} = -2.996$, $p = 0.004$, $d = -0.44$), Medical care *frequency* ($t_{(55)} = -3.889$, $p < 0.001$, $d = -0.56$), Total *frequency* ($t_{(54)} = -4.144$, $p < 0.001$, $d = -0.50$), and Total *difficulty* ($t_{(50)} = -3.104$, $p = 0.003$, $d = -0.46$), with mothers experiencing significant more pediatric parenting stress than fathers on all these scales, with small to medium effect sizes.

Table 2: Means and standard deviations of fathers and mothers on the subscales and total scales of pediatric parenting stress at the first measurement occasion.

Pediatric parenting stress	Fathers (n=56) M (SD)	Mothers (n=56) M (SD)	*t*	*p*	*d*
Communication frequency	17.7 (5.13)	20.2 (4.78)	*-3.605*	*.001*	*-0.50*
Communication difficulty	14.1 (4.06)	16.4 (4.85)	*-3.997*	*<.001*	*-0.51*
Emotional distress frequency	28.4 (7.70)	32.5 (8.42)	*-3.994*	*<.001*	*-0.51*
Emotional distress difficulty	29.6 (9.97)	34.1 (10.51)	*-2.996*	*.004*	*-0.44*
Medical care frequency	18.4 (5.19)	21.3 (5.20)	*-3.889*	*<.001*	*-0.56*
Medical care difficulty	11.5 (4.28)	12.8 (4.01)	*-1.950*	*.057*	*-0.32*
Role functioning frequency	18.4 (5.04)	19.0 (4.54)	*-1.082*	*.284*	*-0.13*
Role functioning difficulty	16.9 (5.23)	18.5 (6.56)	*-1.681*	*.099*	*-0.27*
Total frequency	83.1 (20.03)	93.0 (19.65)	*-4.144*	*<.001*	*-0.50*
Total difficulty	71.4 (20.42)	81.4 (22.84)	*-3.104*	*.003*	*-0.46*

Pediatric parenting stress over a one year period

Table 3 summarizes mean scores and standard deviations on the subscales and total scales assessing pediatric parenting stress of the fathers and mothers who completed both measurement waves.

Fathers reported significantly more perceived pediatric parenting stress over time on the Emotional distress *difficulty* ($t_{(28)}$ = -2.132, *p = 0.042, d* = -0.32) and Total *difficulty* ($t_{(26)}$ = -2.345, *p = 0.027, d* = -0.29) scales, with small effect sizes. However, they reported significantly less pediatric parenting stress on the Medical care *frequency* scale ($t_{(30)}$ = 2.861, *p = 0.008, d* = 0.10) over time, with a small effect size.

Mothers reported significantly less perceived pediatric parenting stress over time on the Emotional distress *frequency* ($t_{(42)}$ = 2.403, *p = 0.021, d* = 0.31), Medical care *frequency* ($t_{(43)}$ = 3.513, *p = 0.001, d* = 0.66), and Total *frequency* ($t_{(42)}$ = 2.263, *p = 0.029, d* = 0.37) scales, with small to medium effect sizes.

Although mothers reported to experience higher levels of pediatric parenting stress than fathers on almost all subscales at the second measurement occasion, differences were not statistically significant.

Table 3: Means and standard deviations of fathers and mothers on the subscales and total scales of pediatric parenting stress over time.

Pediatric parenting stress	Fathers (n=31)					Mothers (n=44)				
	Time 1 M (SD)	Time 2 M (SD)	*t*	*p*	*d*	Time 1 M (SD)	Time 2 M (SD)	*t*	*p*	*d*
Communication F	16.6 (4.7)	17.2 (4.3)	*-0.76*	*.451*	*-0.13*	19.6 (4.9)	19.1 (4.9)	*0.59*	*.559*	*0.10*
Communication D	13.2 (3.6)	14.6 (4.4)	*-1.60*	*.121*	*-0.32*	15.9 (4.6)	15.3 (4.5)	*0.78*	*.442*	*0.13*
Emotional distress F	26.0 (7.0)	28.5 (7.9)	*-1.67*	*.105*	*-0.27*	32.0 (8.5)	29.4 (8.3)	*2.40*	*.021*	*0.31*
Emotional distress D	28.0 (9.6)	31.4 (11.9)	*-2.13*	*.042*	*-0.32*	33.4 (10.6)	31.5 (11.5)	*1.17*	*.250*	*0.17*
Medical care F	17.7 (5.2)	17.2 (4.3)	*2.86*	*.008*	*0.10*	20.8 (5.6)	17.4 (4.7)	*3.51*	*.001*	*0.66*
Medical care D	11.7 (4.9)	12.5 (5.0)	*-0.79*	*.435*	*-0.16*	12.9 (4.4)	11.8 (4.6)	*1.44*	*.158*	*0.24*
Role functioning F	17.6 (4.4)	17.8 (4.3)	*-0.35*	*.731*	*-0.05*	18.9 (5.1)	18.1 (4.7)	*0.98*	*.332*	*0.16*
Role functioning D	17.0 (5.4)	17.4 (5.8)	*-0.47*	*.640*	*-0.07*	17.6 (6.1)	17.2 (5.7)	*0.47*	*.640*	*0.07*
Total F	77.8 (18.9)	79.2 (17.5)	*-0.52*	*.610*	*-0.08*	91.2 (21.1)	83.9 (19.4)	*2.26*	*.029*	*0.37*
Total D	68.1 (19.8)	74.3 (22.6)	*-2.35*	*.027*	*-0.29*	79.0 (22.6)	74.6 (22.9)	*1.21*	*.233*	*0.19*

F = frequency
D = difficulty

Discussion

Although previous studies showed that parents of children with T1DM experience parenting stress [3, 20], especially parents of young children [4-6], information about differences in pediatric parenting stress between fathers and mothers of young children with T1DM and changes in pediatric parenting stress over time was lacking. This information, however, is needed to know whether pediatric parenting stress differs between fathers and mothers and whether the pediatric parenting stress increases or decreases over time. With this information, health care providers are able to provide the support and help these parents might need. Therefore, the present study examined whether the amount of perceived pediatric parenting stress of fathers and mothers of young children (0-7 years) with T1DM differed and changed over time.

In line with research in other pediatric samples [18, 21, 22] and studies including fathers and mothers of older children with T1DM [8, 10], we also found that mothers reported higher levels of pediatric parenting stress than fathers. Mothers of children with T1DM are generally known to be the primary caretaker of their children [16], and therefore take on the responsibility of the diabetes management of their child [15], which could indicate higher levels of pediatric parenting stress in mothers than in fathers. Fathers, on the other hand, are generally known to be more involved in playful activities with their children [23], and more responsible for household tasks when having a child with T1DM [24], which could indicate less pediatric parenting stress in fathers than in mothers. With this information, health care providers should try to involve fathers as much as possible in the diabetes treatment. When fathers become aware of the fact that mothers, on average, perceive more pediatric parenting stress, these fathers might become more involved in the treatment of their child and may thereby lower the pediatric parenting stress of their spouses. Furthermore, it is also important to get fathers more involved in diabetes treatment, as previous research has shown that paternal involvement in disease management is associated with better marital, maternal, and family outcomes in families with children with a chronic disease [25].

The mothers still reported higher levels of pediatric parenting stress than fathers on most scales at the second measurement occasion; however, the differences were not significant anymore. Therefore, the results could indicate that mothers get more used to the treatment demands and accompanying stress and therefore report less stress over time. It could also indicate that families that participated shifted more responsibilities from the mothers to the fathers, which could raise the difficulty in pediatric parenting stress of the fathers and decrease the frequency of pediatric parenting stress of the mothers. These possible explanations are supported by the results as fathers reported a significant

increase in emotional distress and total score, whereas mothers reported a significant decrease in these same scales. Furthermore, both fathers and mothers reported to perceive a significant decline in the medical care frequency subscale, which might indicate that parents get more used to the demanding treatment regimen of their child. It could also mean that the children themselves get more responsibility in their medical care, and, therefore, parents report less stress in the medical care subscale. However, as the age of the children in our sample was between 0-7 years and diabetes care responsibilities are gradually transferred to the child in the preadolescent years [26], the first explanation might be more probable.

The present study has some limitations, for example, we could only include 31 fathers (55%) and 44 mothers (79%) when examining the perceived pediatric parenting stress of fathers and mothers over time. Furthermore, despite contacting the families by mail or phone, not all parents completed the second measurement wave. Reasons for not participating were: too busy, not interested, no reason was given, or we could not get in contact with them by phone. Furthermore, the sample consisted of almost only Caucasian participants (97%) and the educational level of participating parents was relatively high, therefore, the results may not be generalizable to parents with other ethnic backgrounds and/or a lower educational level. Also, most fathers and mothers (96%) in our sample were cohabiting, married or registered partners. It is likely, however, that single fathers and mothers take on more caretaking activities and therefore could experience more pediatric parenting stress than married fathers and mothers. Therefore, the findings are also not generalizable to all families, especially in single-parent-families. Furthermore, we tested the differences in pediatric parenting stress between fathers and mothers (first measurement occasion) and over time (second measurement occasion) with a p-value of 0.05. We did not correct for multiple tests (for example Bonferroni correction), because the subscales correlated highly with each other and we did not want to lose power. We preferred a type 1 error over a type 2 error as this is the first study examining pediatric parenting stress between fathers and mothers of young children with T1DM and over time, and therefore, we wanted to be sure that all the possible differences were detected for future studies. Although significant, most differences in pediatric parenting stress between fathers and mothers were rather small, for example, a difference of 2.3 – 2.5 points on the communication scale which could range from 9-45 points. This was reflected in the small effect sizes. Future research should examine whether these differences are also clinically relevant, by asking the mothers whether and how the amount of stress influences their daily lives and examining whether more support from their spouses could lower the stress of the mothers. This was also the case when

examining the perceived pediatric parenting stress over time. For example, fathers reported a change of 0.5 points in the medical care subscale, with a small effect size (0.10). Although significant, future research should examine whether this (and other) differences are also clinically relevant, by asking the fathers whether they experience more stress than before and whether the higher levels of stress have an influence on their lives.

Research in teens [27] and young children with T1DM [28], have shown that higher levels of pediatric parenting stress are associated with lower (diabetes-specific) quality of life of the children. If health care providers are able to assist with lowering the levels of pediatric parenting stress, by for example changing treatment regimen from multiple daily insulin injections to pump therapy [4] or proving a support program for parents [13], they could enhance the (diabetes-specific) quality of life of the children, which is an important treatment goal [29]. Future research should focus on the development or evaluation of these or similar interventions.

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Chapter 7

Attachment as a protective factor in the relationship between pediatric parenting stress and child outcomes in young children with type 1 diabetes

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Submitted

ABSTRACT

Background
Child behavioral problems, a (sub)optimal HbA_{1c}, and lower child quality of life (QoL) can contribute to increased parenting stress among fathers and mothers of children with type 1 diabetes (T1DM). However, the quality of the relationship between parent and child might form a buffer in the association between these factors and parental stress. Therefore, we aim to examine whether the quality of the attachment relationship moderates the relationship between child outcomes (behavioral problems, HbA_{1c} level, and diabetes-specific QoL (DSQoL)) and pediatric parenting stress in parents of young children (0-7 years) with T1DM.

Methods
77 families of young children with T1DM (0-7 years) completed questionnaires about the behavioral problems and DSQoL of their child. The quality of the attachment relationship was observed during a home visit and assessed with the Attachment Q-Sort by an independent observer. HbA1c level was extracted from the medical records.

Results
It appeared that attachment moderated the relationship between pediatric parenting stress and child behavior problems (β (interaction) = -0.203, p = 0.078, R^2 change = 0.04). This means that when parents reported higher levels of pediatric parenting stress, the child behavior problems also increased although not as much as when the child was securely attached to his/her parent. Furthermore, attachment also moderated the relationship between pediatric parenting stress and DSQoL (β (interaction) = 0.201, p = 0.075, R^2 change = 0.037). This means that when parents report higher levels of pediatric parenting stress, the DSQoL decreases although not as much as when the child is securely attached to his/her parent.

Conclusion
Pediatric diabetes practice should focus on the attachment relationship between parent and child, as this appears to be a protective factor for behavioral problems and DSQoL in children with T1DM; when these children are securely attached, they are less vulnerable to high levels of pediatric parenting stress and, therefore, experience less behavior problems and a higher DSQoL.

Introduction

When children are diagnosed with type 1 diabetes mellitus (T1DM) at an early age, their parents get full responsibility for the diabetes management of their child with support from the medical pediatric team. This is a challenging task, especially when the children are too young to adequately deal with the disease themselves [1, 2], because the parents have to monitor the blood glucose several times each day, administer insulin, regulate food intake, and monitor these parameters in conjunction with the level of physical activity of their child, 24 hours/day, 7 days/week . This responsibility of constant care can lead to elevated levels of parenting stress in these families [3-5].

Various factors in children with T1DM have been associated with increased parenting stress, including child behavior problems, (sub)optimal glycemic control (HbA_{1c} level), and lower child quality of life (QoL) [6, 7]. Parents who experienced high levels of parenting stress, reported more child behavior problems than parents with low levels of parenting stress [8-10]. Parenting stress is also associated with suboptimal HbA_{1c} levels in older children and adolescents [6, 7], but this association was not found for young children with T1DM [11]. The relationship between parenting stress and health outcomes is still unclear. Some studies argue that parenting stress might contribute to poor health outcomes in children with T1DM [6-10], whereas other studies found evidence that in young children, high levels of parenting stress were associated with more optimal HbA_{1c} levels [5] or not even related at all [11]. Research regarding the associations between parenting stress and QoL in (young) children and adolescents with T1DM is scarce. Using PubMed, we only found one study, reporting that higher levels of parenting stress were associated with a lower diabetes-QoL of the child [12] which is in line with the results of another study from our research group study [11].

The aforementioned studies about factors associated with parenting stress suggest that higher levels of parenting stress are apparently not beneficial for the children's behavior and QoL, as these children show more child behavior problems and a lower QoL [8-12]. It is of course also possible that the behavior problems and lower QoL of the children induce parental stress. Although more parenting stress was found to be beneficial for more optimal HbA_{1c} levels in younger children [5], however, our research group did not found a correlation between parenting stress and HbA_{1c} levels in young children [11].

A factor that might be important in explaining the relationship between parenting stress and child outcomes is the quality of the "attachment relationship" between parent and child. Attachment is described as the emotional bond between infant and parents [13] that develops over time [14]. Infants form an attachment relationship with their parent, who functions as a secure base for the child to explore the environment [15]. This attachment relationship between

parent and child can be classified as 'secure' or 'insecure' [16]. When children are securely attached, they trust their parents and can rely on them when they need their help, despite their parents' high levels of stress [16, 17]. Therefore, securely attached children will be better able to deal with their parents' stress (and with stress in general) than insecurely attached children [17]. The ability to deal with parent's stress might also affect the number of behavioral problems, the HbA_{1c} level and QoL of the child. A study by Tharner and colleagues [17] found that a secure attachment relationship moderated the effect of parenting stress on child behavior problems in healthy children. Parents of young children with T1DM do not only experience general parenting stress, but also worry about their child's treatment and health, i.e. pediatric parenting stress. Therefore, we aim to examine whether attachment also moderates the relationship between pediatric parenting stress and child outcomes in young children (0-7 years) with T1DM. In particular, the present study will examine the following three questions (see Figure 1):

1. Does attachment moderate the relationship between pediatric parenting stress and behavioral problems?
2. Does attachment moderate the relationship between pediatric parenting stress and glycemic control (HbA_{1c} level)?
3. Does attachment moderate the relationship between pediatric parenting stress and diabetes-specific QoL?

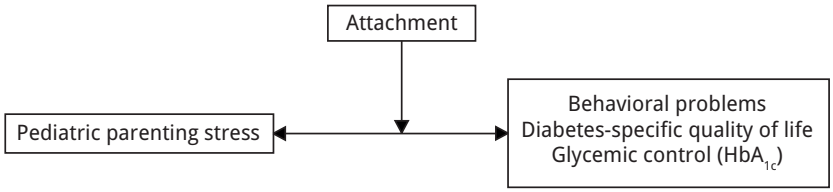

Figure 1: Hypothesized relationships between pediatric parenting stress, attachment, and three child outcomes (child behavior problems, diabetes-specific quality of life and glycemic control).

Methods

Patients and procedure

All children with T1DM (aged 0-7 years) and their parents were recruited from 15 hospitals/institutions in the middle and southern part of the Netherlands (including Diabeter, a national center for pediatric and adolescent diabetes care and research, and Kidz&Ko, a partnership between seven pediatric diabetes clinics). Parents who lacked basic proficiency in Dutch were excluded, as well as children with Down syndrome and/or other mental disabilities, and children with an Autism Spectrum Disorder (n=17). Of the 121 eligible families, 77 families

(64%) agreed to participate. After a home visit in which parent-child interactions were videotaped (for a detailed description of the procedure of the home visits see Nieuwesteeg et al. 2014 [18]), the quality of the attachment relationship was established by an independent observer who used the Attachment Q-Sort (AQS) instrument (for a description of the AQS, see *measures*). Furthermore, parents were asked to complete a questionnaire assessing (socio)demographic and clinical variables, child behavior problems, and diabetes-specific child QoL. We only assessed the attachment relationship of the parents who were videotaped during the home visit. Of the 77 observed parents, we could compute the total scores of 83% (n=64) of the questionnaires assessing pediatric parenting stress, 86% (n=66) of the questionnaires measuring child behavior problems, and 91% (n=70) of the questionnaires that focused on the diabetes-specific QoL of their child. The study was approved by the Medical Ethical Review board of St. Elisabeth Hospital Tilburg (date: 25-05-2010).

Measures

(Socio)demographic and clinical variables

The parents completed a questionnaire with (socio)demographic and clinical characteristics including, gender and age of the child; relationship with the child (father or mother), marital status (single, cohabiting, or married/registered partners), educational level (primary education, 12 years of formal education, 15-16 years of formal education, or other), treatment regimen of the child (insulin pump or multiple daily insulin injections), and frequency of blood glucose monitoring a day.

Metabolic control

Glycemic control (HbA_{1c} level, measured closest to the home visit) was extracted from the child's medical record at the hospital where the child was treated for T1DM, after receiving written consent from the parents.

Attachment

The Attachment Q-Sort (AQS) [19] was used to assess infant attachment behavior in home settings. The AQS consists of 90 cards, each describing a different child behavior (e.g., "When parent says to follow, child does so", "Child readily shares with parent or let him/her hold thing if he/she asks to"). After the home-visit, the observer (AN) sorted the 90 descriptive statements into nine piles of ten cards each, ranging from very much like the child (pile 9) to very much unlike the child (pile 1). A security score was obtained by correlating the child's Q-sort description with the criterion sort for a prototypically secure infant, provided by experts. Security scores range from +1.00 for a perfectly secure

attached infant to -1.00 for a most insecure attached infant. The observer (AN) was thoroughly trained by the last author (HvB), who has extensive experience in applying the AQS (e.g. [20]).

Pediatric parenting stress

Pediatric parenting stress was assessed with the 42-items Pediatric Inventory for Parents (PIP) [21, 22]. All items of the PIP were scored on a 5-point Likert scale on both *frequency* and *difficulty*. The items of the PIP are aggregated into four subscales: Communication with the child and the medical team, Emotional distress, Medical care and Role functioning. The scale scores were summed to get a total *frequency* and total *difficulty* score. Higher scores reflect more frequency and difficulty in pediatric parenting stress. Adequate internal consistency and construct validity of the original version of the PIP have been reported [21]. In the present study, the score on the total *frequency* scale (α = 0.93) was used to represent the pediatric parenting stress of the participating parents.

Child behavior problems

To assess child behavior problems, the parents completed the Strengths and Difficulties Questionnaire (SDQ) [23]. The SDQ is a brief behavioral screening questionnaire and measures the presence of psychosocial problems and the strengths of the child. The questionnaire consists of 25 items, covering the following five domains: emotional problems, conduct problems, hyperactivity/inattention, peer relationship problems, and pro-social behavior. The 25 items are formulated on the basis of propositions and relate to the past 6 months. Some propositions are oppositely formulated. The four domains assessing problem behavior (emotional problems, conduct problems, hyperactivity/inattention, and peer relationship problems) were summed to get a total problem score (α = 0.80). Higher scores correspond to more problem behavior. Research showed that the Dutch translation of the SDQ has acceptable to good psychometric properties [24].

Quality of life of the children

Diabetes-specific QoL (DSQoL) was measured with a proxy version of the child self-report questionnaire (Smiley Faces) for young children, composed by the Hvidøre Study Group [25]. This questionnaire has been modified in a proxy-report form with permission of the authors, so that the parents can complete the questionnaire for their child. The questionnaire comprises 19 items about feelings of the child in relation with his or her diabetes (for example, items about administering insulin, health, leisure time, and school/nursery/daycare). The

items are rated on a 5-point Likert scale. Scores are (re)coded so that a higher score corresponds to a better DSQoL. All items were summed to get a total DSQoL score (α = 0.86). The originally child self-report questionnaire showed good reliability and validity [25].

Statistical analyses

Statistical analyses were conducted using the Statistical Package for the Social Sciences (SPSS, version 19). Frequencies and descriptive statistics were used to describe the (socio)demographic and clinical characteristics of the participating parents and children. To examine whether the attachment relationship moderates the relationship between pediatric parenting stress and child outcomes (behavioral problems, HbA_{1c} level, and diabetes-specific QoL), we performed hierarchical regression analyses for each outcome measure. First we fitted the model with centered pediatric parenting stress, centered attachment, and their product as predictors. If the moderator was not significant, we refitted the model with main effects only.

Because of the explorative nature of the present study, a modest sample size, and because we did not want to overlook possible moderation effects of attachment (i.e., reduce the risk of a Type II error), we a priori choose a significance level of 0.10 for the statistical tests. This means that we accept a risk of a Type I error that is higher than the conventional 5% level, but we do so to gain power. However, this means that possible significant results should be considered as preliminary evidence of moderation and further research is necessary in larger samples.

Results

Sociodemographic and clinical data

Table 1 summarizes the characteristics of the participating parents and children. In total, 77 primary caretakers (74 mothers and 3 fathers) were observed with their child to assess the quality of attachment relationship. Most of the participating parents were married/registered partners (83% of the mothers and 100% of the fathers). Almost half of the participating mothers (49%) had a higher educational level (i.e., approximately 12 years of formal education), and all fathers (100%) had a Bachelor's or Master's degree (i.e., approximately 15 years of formal education). Their children had a mean age of 5 years (SD= 1.5, range: 2-7 years) 53% were boys. Most children (82%) received pump therapy. On average, parents monitored their child's blood glucose 6 times a day (range: 2-20). The mean HbA_{1c} level of the children was 59 mmol/mol or 7.6% (range 32-80 mmol/mol or 5.1%-9.5%).

Table 1: Sociodemographic and clinical characteristics of participating parents and their young children with type 1 diabetes.

			% (N)	M	SD
Children	Sex	Boys	53% (41)		
		Girls	47% (36)		
	Age (years)	(range 2-7)		5.12	1.52
	HbA_{1c}	(range 32 mmol/mol - 80 mmol/mol)		59	9.03
		(range 5,1% - 9,5%)		7.6	0.8
	Treatment	Insulin pump	82% (63)		
		Multiple daily insulin injections	18% (14)		
	Blood glucose monitoring	Times a day (range 2-20)		6.4	2.5
Parents	Total	Mothers	96% (74)		
		Fathers	4% (3)		
	Marital status (mothers)	Single	7% (5)		
		Cohabiting	13% (10)		
		Married / registered partners	70% (52)		
		Missing	10% (7)		
	Marital status (fathers)	Single	0% (0)		
		Cohabiting	33% (1)		
		Married / registered partners	67% (2)		
	Educational level (mothers)	Primary education only	1% (1)		
		About 12 years of formal education	49% (36)		
		15-16 years of formal education	39% (29)		
		Other	1%(1)		
		Missing	10% (7)		
	Educational level (fathers)	Primary education only	0% (0)		
		About 12 years of formal education	0% (0)		
		15-16 years of formal education	100% (3)		

Attachment as a moderator between pediatric parenting stress and child outcomes

As Table 2 shows, attachment moderated the relationship between pediatric parenting stress and child behavior problems, β = -0.203, $t_{(57)}$ = -1,794, *p = 0.078* (two-tailed), where 4% of the total variance in child behavior problem is uniquely explained by the moderation effect (i.e., R^2 change = 0.04). To better understand the interaction effect between the continuous indicators stress and attachment, we graphically probed the interactions (e.g., [26]) by plotting the estimated simple regression lines between parenting stress and child behavior problems at three selected levels of attachment. The three levels include an attachment score of 1 SD below average (low) average attachment score, (medium), and an attachment score 1 SD above average (high) (see Figure 2). For low levels of attachment, the reported child behavior problems increased with higher levels of pediatric parenting stress, whereas for high levels of attachment the reported child behavior problems increased less with higher levels of pediatric parenting stress. Also, for low levels of stress (1 SD below average), differences between child behavior problems and attachment were negligible. This means that when parents report higher levels of pediatric parenting stress but low levels of attachment, the child behavior problems also increase although not as much as when the child is securely attached to his/her parent.

As Table 2 shows, we found no evidence that attachment moderates the relationship between pediatric parenting stress and HbA_{1c} level (β = -0.023, $t_{(60)}$ = -0,179, *p = 0.858* (two-tailed), R^2 change = 0.01). Regression analysis without the interaction effect also showed no significant results (R^2 = 0.021, p = 0.523). This indicates that both the quality of attachment relation and the level of pediatric parenting stress are unrelated to the HbA_{1c} level of the child in the present study.

Furthermore, as Table 2 shows, attachment moderated the relationship between pediatric parenting stress and DSQoL, β = -0.201, $t_{(60)}$ = 1,810, *p = 0.075* (two-tailed), where almost 4% of the total variance in child behavior problem is uniquely explained by the moderation effect (i.e., R^2 change = 0.037). To better understand the interaction effect between the continuous indicators stress and attachment, we plotted the estimated simple regression lines between parenting stress and DSQoL at three levels of attachment (see Figure 3). For low levels of attachment, the reported DSQoL decreased with higher levels of pediatric parenting stress, whereas for high levels of attachment the reported DSQoL only decreased slightly with higher levels of pediatric parenting stress. Also, for low levels of stress (1 SD below average), differences between child behavior problems were negligible. This means that when parents report higher levels of pediatric parenting stress, the DSQoL decreases although not as much as when the child is securely attached to his/her parent.

Table 2: Hierarchical Regression analyses predicting child outcomes (child behavior problems, HbA_{1c} level, and diabetes-specific quality of life) from attachment, stress, and their interaction.

	Child Outcomes					
	Child behavior problems		HbA_{1c} level		Diabetes-specific quality of life	
Predictor	ΔR	β	ΔR	β	ΔR	β
Step 1: Main effects only	.254		.021		.283	
Attachment		-0.181		0.004		0.119
Stress		0.445***		-0.143		-0.508***
Step 2: Including interaction	.040		.001		.037	
AttachmentxStress		-0.023*		-0.023		0.201*
Total R	.294		.022		.320	
Adjusted Total R	.256		.000		.286	
n	60		63		63	

[1] Fully standardized regression coefficients [27]
[2] Reduction in the R-square when the moderator is excluded from the regression model; this value indicates the proportion of the variance in the outcome measure that is uniquely accounted for by the moderation effect.
* p < .10. ** p < .05. *** p < .001.

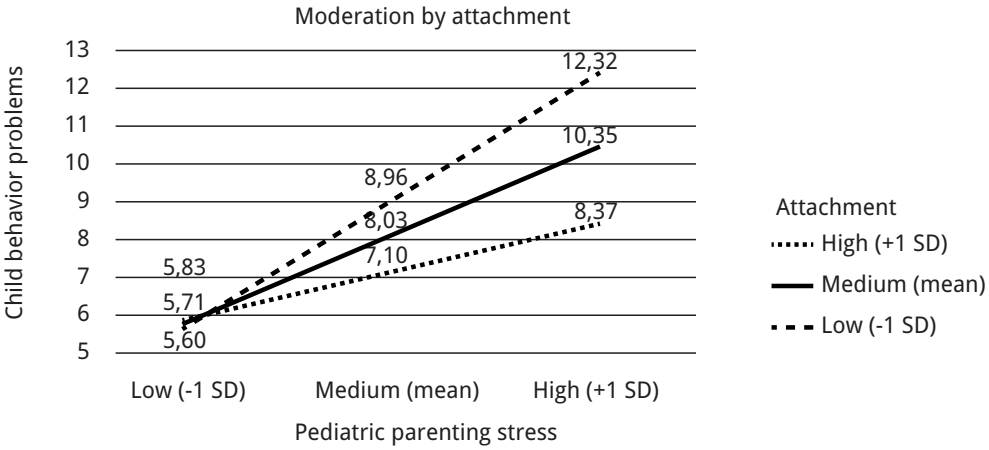

Figure 2: Moderation by attachment on the relationship between pediatric parenting stress and child behavior problems.

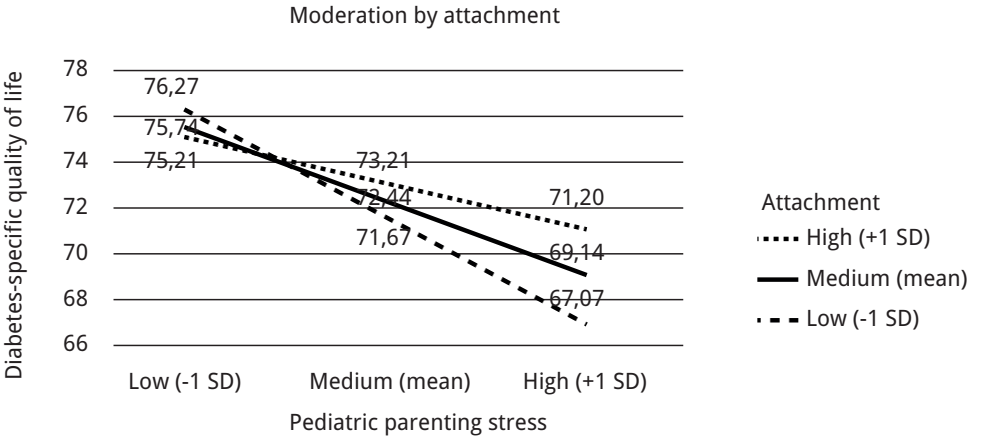

Figure 3: Moderation by attachment on the relationship between pediatric parenting stress and diabetes-specific quality of life.

Discussion

The results of the present study suggest that the quality of the attachment relationship between parent and child moderates the association between pediatric parenting stress and child behavior problems. In particular, in families where children are more securely attached to their parents, the association between pediatric parenting stress and child behavior problems is weaker than in families where children are less securely attached to their parents. This means that a secure attachment relationship between parent and child could be seen as a protective factor for behavioral problems in children with T1DM induced by

parental stress; when these children are securely attached, they are probably less vulnerable to high levels of pediatric parenting stress and, therefore, experience less behavior problems. This is consistent with results from previous research on the moderation effect of attachment on the relationship between parenting stress and child behavior problems in healthy children [17]. Results also suggest that the attachment relationship moderates the association between pediatric parenting stress and DSQoL. Parents who experienced higher levels of stress reported a lower DSQoL of their child. However, when their children were securely attached they reported a better DSQoL in spite their high levels of stress. This result also means that a secure attachment relationship between parent and child could be seen as a protective condition in children with T1DM; when these children are securely attached, they are less affected by high levels of pediatric parenting stress and, therefore, experience a better DSQoL. The quality of the attachment relationship is believed to influence later child development [28, 29] and is therefore an important focus of interventions [13, 30]. We believe that it is important for healthcare providers to give help and support to young patients in creating a secure attachment relationship with their parents. Research has shown that interventions focusing on parental insensitivity are most effective in enhancing the attachment relationship [31]. A way to treat parental insensitivity is video-feedback training [32], in which the parents are confronted with their own behavior. It is important to focus on the parent-child dyad in the treatment of T1DM, as a secure attachment relationship between parent and child could have an influence on the relationship between pediatric parenting stress and child behavior problems / DSQoL. Future research should therefore focus on the development of interventions to improve the attachment relationship between parents and young children with T1DM as a secure attachment relationship could reduce child behavior problems and improve the DSQoL, especially in families with parents experiencing high levels of pediatric parenting stress.

Unlike a moderation effect of attachment on the relationship between pediatric parenting stress and child behavior problems/DSQoL, results suggest that attachment does not act as a moderator on the relationship between pediatric parenting stress and HbA_{1c} level. Hence, we have no evidence that a secure attachment relationship is a protective factor on the relationship between pediatric parenting stress and the HbA_{1c} level of the child. Research with adults [33, 34] and adolescents [35] with T1DM have found that a less secure attachment relationship predicts suboptimal HbA_{1c} levels and a more secure attachment relationship predicts a more optimal glycemic control. The results of these studies [33-35] might suggest that insecurely attached adolescents are less able to interact adaptively and rely more on other people, for example, clinicians [34]. The studies on attachment in patients with T1DM did find a relationship

between attachment and HbA_{1c} level, however, they rely on questionnaires assessing self- or mother reported attachment [33-35]. We believe that assessing attachment by an independent observer provides more objective information than when using self-reports by parents or adolescents who are perhaps more prone to give socially desirable responses or are influenced in their attachment ratings by factors as infant irritability and parental stress [36]. Because of their age, upcoming independence and their peers, adolescents might not always have a good relationship with their parents, and therefore might report a less secure attachment style, while in fact the attachment relationship is secure. On the other hand, mothers might think their attachment relationship with their child is highly secure, while in fact the attachment relationship is less secure. Or mothers might fill out the questionnaire in a socially desirable way. Therefore, the results of the studies [33-35] showing a relationship between attachment and HbA_{1c} level might be less weaker or even absent, like in our study.

Unfortunately, the present study has a rather small sample size (n=77). Because of the small sample, which may limit the power, and the explorative nature of the present study, we choose a significance level of 0.10. We were aware of a higher risk of a Type 1 error (finding a significant result while in fact the result is not significant), but given that possible moderators may have important implications for designing interventions in the clinical practice, we did not want to miss any possible moderation effects due to lack of power at this stage of the research. We think a Type 1 error of 10% is acceptable given this is an exploratory study. Future research is necessary in larger samples in order to confirm whether attachment indeed acts as a moderator on the relationship between pediatric parenting stress and 1) child behavior problems and 2) DSQoL. Furthermore, our sample consisted of mostly Caucasian and higher educated parents, which is not representative of the population in the Netherlands [37].

Despite the limitations of the present study, we would like to highlight the importance of a secure attachment relationship. The results suggest that a secure attachment relationship might act as a protective factor on the relationship between child behavior problems and DSQoL, which are important child outcomes. Therefore, clinicians should focus more on the relationship between parent and child or should involve psychologist more often in order to improve child behavior and DSQoL. Fortunately, clinicians become more and more aware of the psychological aspects of T1DM, but it is important to not only focus on the child itself, but also on the parent-child relationship.

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Chapter 8

Summary and general discussion

The present thesis has a focus on the parent-child interactions, parenting stress and child outcomes in families with a young child (0-7 years) with type 1 diabetes (T1DM). As described in the general introduction of this thesis (**Chapter 1**), T1DM is a common chronic disease in pediatric care settings [1-4]. Research in the youngest patient group, however, has remained scarce, although the diagnosis of T1DM and having T1DM has a large impact on the lives of both the children themselves and their parents, and the way they interact with each other. The lack of knowledge about the exact impact of T1DM on families with the young patients with diabetes prompted us to formulate the following research aims: to gain more insight into the impact of T1DM in families with a young child (0-7 years) with T1DM, with a specific focus on the quality of parent-child interactions, the level of general and pediatric parenting stress, and on child outcomes (HbA_{1c} level, quality of life (QoL), and child behavior problems), and also to study the potential buffering role of attachment. In this summary and general discussion chapter, an overview of the main findings will be given and discussed, as well as practical implications, possible future directions and an overall conclusion.

Overview of the main findings

Quality of life of children with T1DM

Nowadays, most healthcare providers do not only focus on optimizing the HbA_{1c} levels of children with T1DM, but also on guarding or enhancing their QoL where possible [5]. At the start of the present PhD-trajectory, a systematic review about the QoL in children with T1DM was lacking. Therefore, we conducted a systematic review to give an overview of the QoL of children and adolescents with T1DM (**Chapter 2**). More specifically, we reviewed 17 studies [6-22] examining the generic QoL of both children with T1DM and their healthy peers. Although some studies reported a lower QoL in children with T1DM [6, 10, 11, 16-20], others reported mixed results [12-15, 21] and some even reported a better QoL for children and adolescents with T1DM compared to healthy peers [7-9, 22]. On average, when taking samples sizes into account, it appeared that children and adolescents with T1DM showed no differences in physical, psychosocial, and overall QoL compared to their healthy peers. Diabetes-specific problems, like worrying about their disease, feeling like a burden on their family, and being less satisfied with their social life, however, were certainly present. When examining differences between boys and girls with T1DM regarding their (diabetes-specific) QoL, we found that girls reported a lower QoL than boys. No clear pattern was found when comparing the (diabetes-specific) QoL of children with T1DM across different developmental stages. The systematic review further revealed that studies assessing (diabetes-specific) QoL of young children (< 8 years) and studies comparing different age groups were still limited. Furthermore, the review also

showed that longitudinal studies examining the course of (diabetes-specific) QoL were still lacking and form an important gap in the literature.

Developing an instrument to measure the quality of parent-child interactions

Because parents are fully responsible for the diabetes-management of their young children [23], diabetes-management tasks could influence the interactions between parent and child and vice versa. However, only a few studies had examined the quality of parent-child interactions in young children with T1DM [24-29]. The studies that had been conducted have important limitations. For example, they used observation methods that are time consuming and in which behaviors cannot be evaluated in the context of other behaviors [30-32]. Therefore, we developed a time efficient observation method with rating scales, specifically designed to assess the quality of parent-child interactions in diabetes-specific situations in a valid and detailed way. In **Chapter 3** we have described the initial plan to develop this new observation instrument in a sample of approximately 120 young children with T1DM and their parents. How the development of the OKI-DO (Ouder Kind Interactie – Diabetes Onderzoek, i.e. Parent Child Interaction Diabetes Research) observation instrument actually took place, is described in **Chapter 4** (see Figure 1).

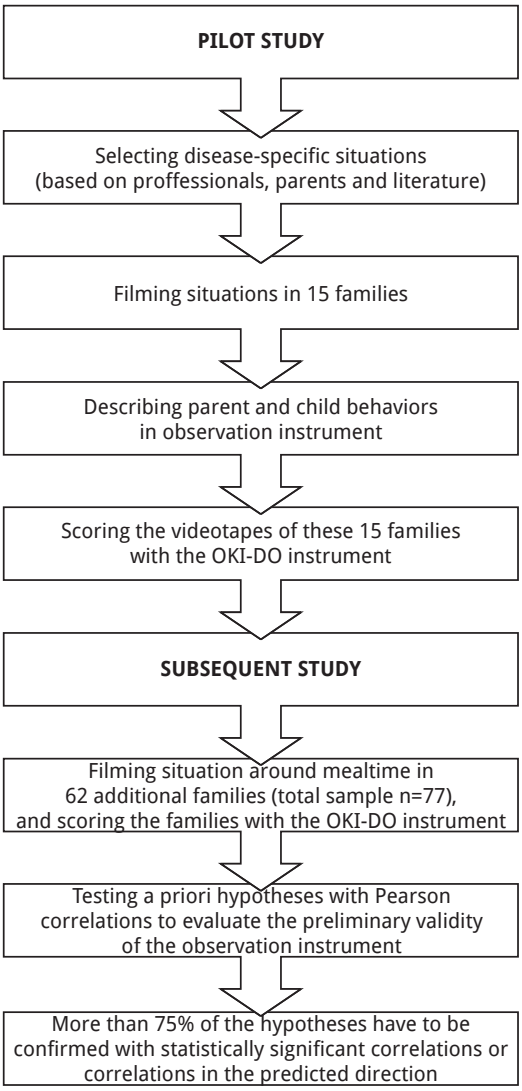

Figure 1: Constructing the OKI-DO observation instrument.

At first, we needed to determine which situations were the most relevant to observe. Based on literature and interviews with experts, it became clear that the mealtime (including glucose monitoring and insulin administration) was the most salient situation. Therefore, we filmed the mealtime situation in 15 families with a young child with T1DM. All parent and child behaviors were described in four parent domains ("*emotional involvement*", "*limit setting*", "*respect for autonomy*", and "*quality of instruction*"), four child domains ("*negative behavior*", "*avoidance*", "*cooperative behavior*", and "*child's response to injection*") and two

family domains ("*emphasis on diabetes*" and "*mealtime structure*") in the OKI-DO instrument. After some minor improvements, the new observation method was tested in these 15 families. This small pilot study resulted in a usable and reliable (the weighted kappa of 0.73 indicated a good inter-rater reliability) observation instrument to assess parent-child interactions during mealtime in families with a young child with T1DM. After this, the observation instrument was tested in a larger study in order to rigorously evaluate the validity of the instrument. Testing Pearson correlations between the OKI-DO instrument and a generic observation [33] during free play and a child behavior questionnaire [34], showed encouraging support for the preliminary validity of the OKI-DO instrument. In future research, the validity of the OKI-DO instrument should be further examined as our sample consisted of rather high functioning families with homogeneous (ethnic) backgrounds and educational levels of parents.

Associations between parent-child interactions and child outcomes

The development of the OKI-DO instrument has enabled us to investigate the associations between the quality of parent-child interactions and health outcomes; this study is described in **Chapter 5**. The results showed that parents who were more emotionally involved had children with more suboptimal HbA_{1c} levels, which is in accordance with a study including older children with T1DM [35]. However, the direction of this association between suboptimal HbA_{1c} levels and parental emotional involvement remains unclear. It seems plausible that suboptimal HbA_{1c} levels caused parents to be more emotionally involved in trying to delay the onset of complications [35]. At the same time, parents who are more emotionally involved could deliberately set higher HbA_{1c} targets as they fear hypoglycemic events [36]. Furthermore, it appeared that children with suboptimal HbA_{1c} levels expressed more discomfort (like tightening their eyes or resisting) during glucose monitoring / insulin administration, which might be due to needle-phobia, a low pain threshold, or more emphasis on the disease because of a suboptimal HbA_{1c} level. We did not find any significant correlations between the quality of parent-child interaction and (diabetes-specific) QoL of the children. This could be due to the fact that we used observations instead of self-report questionnaires on the quality of parent-child interaction. Research in youth with T1DM has shown that self-report parent-child interactions did correlate with the QoL of the child, while observed parent-child interactions did not [37]. Parents using 'negative' interactions, might be more negative in general and therefore also view the QoL of their child worse than it actually is. In the same way, parents with positive interaction patterns might be more positive in general and therefore view the QoL of their child better than it actually is. Using direct observations may reflect a more objective view than self-report questionnaires [38].

Parenting stress in fathers and mothers of young children with T1DM

Parents are faced with a rather complex treatment regimen of their child with T1DM: they have to monitor the blood glucose level of their child several times a day, regulate their food intake, and administer insulin. This has to be done 24 hours a day. Therefore, it is not surprising that parents of children with T1DM experience stress. Research has shown that parenting young children with T1DM, indeed, can be stressful [39-41]. Furthermore, in our study, we also examined associations between (pediatric) parenting stress and health outcomes (see **Chapter 5**). Although we found no significant association between (pediatric) parenting stress and HbA_{1c} level of the child, the results showed that both general and pediatric parenting stress were significantly correlated with the generic QoL (r ranged between -0.40 to -0.56) and diabetes-specific QoL (r ranged between -0.39 to -0.70) of the children. As (pediatric) parenting stress is related with the generic and diabetes-specific QoL of the child, a randomized controlled trail should examine whether it is useful for health care providers to closely monitor the (pediatric) parenting stress when treating children with T1DM, in order to improve the (diabetes-specific) QoL of the children and/or lower the (pediatric) parenting stress.

When examining pediatric parenting stress in parents of young children with T1DM, fathers are generally being overlooked [39]. It is important to have information about the pediatric parenting stress of fathers as the involvement of fathers in families with a child with T1DM is associated with better family, maternal and marital family outcomes [42]. Unfortunately, research on pediatric parenting stress which included both fathers and mothers of young children with T1DM was lacking. Therefore, we have examined the perceived pediatric parenting stress of both fathers and mothers of young children with T1DM in **Chapter 6**. In line with research in parents of older children with T1DM [43, 44], the results showed that fathers reported (significantly) less pediatric parenting stress than mothers (d ranged between -0.13 to -0.56). Furthermore, we also examined the course of this pediatric parenting stress in both fathers and mothers, to examine whether pediatric parenting stress changes over time. On average, it appeared that mothers reported (significantly) less pediatric parenting stress, while the fathers reported (significantly) more pediatric parenting stress over a one year period. This could indicate that the mothers, who are generally known to be more responsible for the caretaking of their children [45], shift more responsibilities toward the fathers after a while as they get more used to the demands of the diabetes-management of their child. Therefore, if it is not the case already, fathers should be more involved in the diabetes treatment, as they could share the pediatric parenting stressors with their spouses and improve their family outcomes [42].

Attachment as a buffer in the relationship between pediatric parenting stress and child outcomes

From other studies it appeared that factors such as child behavior problems, HbA_{1c} level and QoL were associated with higher levels of parenting stress in families with a child or adolescent with T1DM [39, 46]. A factor that might be important in explaining the relationship between parenting stress and child outcomes is the quality of the attachment relationship between parent and child. Research in healthy children has found that a secure attachment relationship moderates the negative effect of high levels of parenting stress on child behavior problems [47]. A possible explanation could be that securely attached children are better able to deal with parental stress than insecurely attached children [47]. We examined whether attachment also acted as a moderator on the relationship between pediatric parenting stress and child outcomes (child behavior problems, HbA_{1c} level, and diabetes-specific QoL) in young children with T1DM (see **Chapter 7**). Contrary to our expectations and research with adults [48, 49] and adolescents [50] with T1DM, it appeared that attachment did not moderate the relationship between pediatric parenting stress and HbA_{1c} level. In young children, the parents are still responsible for the diabetes treatment, unlike adolescents and adults who are responsible themselves. Therefore, it could be that research with adults [48, 49] and adolescents [50] showed that a less secure attachment relationship predicts suboptimal HbA_{1c} levels, because these insecurely attached adolescents and adults are less able to interact adaptively and rely more on other people, like clinicians [49] and are, therefore, less able to control their HbA_{1c} level themselves.

However, it was found that a secure attachment relationship did moderate the relationship between pediatric parenting stress and child behavior problems and pediatric parenting stress and diabetes-specific QoL. The results of our study underline the importance of a secure attachment relationship between parent and child: when children were securely attached, they were less affected by high levels of pediatric parenting stress and, perhaps therefore experienced less behavior problems and had a better diabetes-specific QoL. Improving the attachment relationship could reduce child behavior problems and improve the diabetes-specific QoL in families who experience high levels of pediatric parenting stress. Therefore, we believe it is important to provide interventions aimed at optimizing the attachment relationship between parent and young child with T1DM. Research has shown that a focus on improving parental sensitivity is the most effective way to improve the attachment relationship between parent and child [51]. Video-feedback training could be a good intervention method to improve parental insensitivity [52].

Strengths and limitations of the study

Sample

Although we were able to include 15 hospitals instead of the 8 hospitals we initially wanted to include and although the study had a response rate of 64%, we were still not able to include the 120 young children we originally planned to include. This was mainly due to the fact that the participating hospitals had overestimated the number of young children with T1DM that was under treatment in their clinics. Most of the times, the number of children was half of the number that was estimated. Because a national Dutch Type 1 diabetes registry is still lacking, we did not know exactly how many young children are diagnosed with T1DM, but current estimations state a number of 750 [53]. Therefore, with the 77 families in our study, we included about 10% of all young children with T1DM in the Netherlands, which can be regarded as a strength of the study.

The parents that did participate in our study were almost all Caucasian (97%), had rather high educational levels, were mainly two-parent families, and most children received pump therapy (82%). Therefore, we have to be cautious to generalize the findings of our study to families with other ethnic backgrounds, lower educational levels, single-parent families and children on multiple daily insulin injections. Furthermore, the families in our sample on average had a rather high level of functioning with regard to their parent-child interactions, given the low scores on the OKI-DO instrument on '*negative behavior*', '*avoidance*', '*response to injection*', and high scores on '*limit setting*', '*respect for autonomy*', and '*cooperative behavior*'. These high levels of functioning could be a result of the high educational level of the parents: a high educational level is associated with the quality of parent-child interactions [54] and parenting strategies [55, 56]. It could also be that parents who encountered difficulties in the parent-child interactions during diabetes-specific situations refused to participate in the OKI-DO study.

Measurements

By using an observational rating scale to assess the quality of parent-child interactions, we were able to take into account affective components. Studies that use observational methods are probably more accurate than studies that rely on counting behaviors [31, 32, 57]. Furthermore, this way of observing is more time efficient than other observation instruments [30, 31] and we can provide more objective data than when parents were interviewed or when parents report in questionnaires about their quality of parent-child interactions [38]. Although the OKI-DO observation instrument is only preliminary tested with regards to its reliability and validity, the results were promising with more than 90% of the a priori hypotheses confirmed. Moreover, the OKI-DO instrument is the only

observational rating scale that was validated. The validity of other widely used and accepted observation instruments is not examined and need rigorous testing [58].

We used several parent- and self-report questionnaires in the present study to assess child behavior problems, (diabetes-specific) QoL, and (pediatric) parenting stress. Self-report questionnaires are used frequently in psychosocial studies to report on, for example, pediatric parenting stress [39]. However, parent- and self-report questionnaires could inflict response biases like socially desirable responding or acquiescent responding [59] which could have influenced the results. We have tried to minimize response biases by sending the parents the questionnaire before the home-visit, so they could complete them separately without the presence of the researcher or partner and without a time limit. Furthermore, we used the short, 17 item version of the NOSIK [60] to assess general parenting stress. We decided to use this questionnaire because it shows promising reliability and validity [60] and we did not want to burden the parents with a large number of very elaborate questionnaires.

Although it is recommended to assess QoL always from the patients' perspective [61-63], parents usually serve as proxies for children under the age of seven to nine years [64, 65]. Because of the young age of the children in our sample, we have chosen to use proxy-reports for examining the (diabetes-specific) QoL of the children. When examining the generic QoL, we used the TAPQOL (0-5 years) [66] or TACQOL (6-7 years) [67] questionnaire, depending on the child's age. These questionnaires consist of different subscales; therefore, comparing the generic QoL in these different age groups was unfortunately impossible, as from our review it appeared that this information was lacking in the current literature we would have liked to compare the QoL in these different age groups.

Practical implications and possible future directions

Although from our systematic review (**Chapter 2**) it appears that, on average, children with T1DM report a similar QoL compared to their healthy peers, diabetes-specific problems seem to be certainly present. Furthermore, the results showed that girls reported lower QoL compared to boys, which has also been found in other patients groups and healthy children [68, 69]. Assessing and discussing the QoL of adolescents with T1DM periodically has proven to be beneficial [70]. Therefore, healthcare providers should be monitoring the (diabetes-specific) QoL of their patients with T1DM regularly, especially in girls, in order to be able to anticipate on possible problems. Research has shown that monitoring QoL by using electronic patient-reported outcomes (ePROs) contributes to communicate about psychosocial issues in a positive way [71]. We encourage health care providers to periodically assess and discuss the (diabetes-

specific) QoL, for example by using ePROs, of all their patients, including the youngest patients. In young children, parents can serve as proxies [65] or a smiley faces instrument can be used [72] to assess and discuss the QoL of their children. Research on the QoL in this young patient group is scarce and longitudinal studies are needed to be able to say something about the developmental course of (diabetes-specific) QoL in this group. With the information of possible changes in (diabetes-specific) QoL, the patients could be referred to specific health care providers in order to enhance their (diabetes-specific) QoL, e.g. dieticians, psychologists, social workers.

With the development of the OKI-DO observation instrument (**Chapter 3** and **Chapter 4**) health care providers with experience in observing behaviors now have a tool that enables them to quantify parent-child interactions during diabetes-specific situations (mealtime, including glucose monitoring and insulin administration). The OKI-DO instrument makes it possible to determine, for example, whether interventions to improve the quality of the parent-child interactions (like Video Interaction Guidance (VIG)) would be meaningful for families experiencing difficulties with their young child with T1DM. VIG consists of edited video feedback with which a nurse or pedagogic worker can help families in achieving their goals, identifying their strengths and decreasing their weaknesses [73]. For example, VIG in combination with the OKI-DO rating scales could be used to decrease the injection distress of children or intrusiveness of parents in families where this is a problem. VIG is known to be an effective intervention for increasing positive behavior in parents [74, 75]. Therefore, we recommend using the OKI-DO instrument in VIG interventions in families with a young child with T1DM experiencing difficulties during mealtime (including glucose monitoring and insulin administration). When health care providers are able to detect possible problems in an early stage and have the resources to solve and/or overcome these problems, bigger problems later in childhood or adolescence might be avoided.

Because the parents in our study were mostly Caucasian (97%), had higher educational levels than in the Dutch population [76], and the majority of the children received pump therapy (82%), we recommend that future research should include parents with a different ethnic background, a lower educational level and children who are on multiple daily insulin injections, in order to further validate the OKI-DO instrument. Furthermore, test-retest reliability could be examined in future research to further examine the reliability of the observational tool. If the OKI-DO instrument would be used in a larger sample with more younger children, the Child Behavior CheckList [77] could be used to examine the construct validity.

It appeared that more emotional involvement of parents and more expression of discomfort during glucose monitoring were related with suboptimal HbA_{1c} levels (**Chapter 5**). The direction of this relationships remains unclear, but health care providers should keep in mind that the emotional involvement of parents, fear of hypoglycemia, needle-phobia or an emphasize on the disease might play a role in these associations. Furthermore, it appeared that the general and pediatric parenting stress were highly correlated with the (diabetes-specific) QoL of the children in our study (**Chapter 5**). Although the causal relationship here too remains unclear, it is important for health care providers to know that the stress level of parents is related to the (diabetes-specific) QoL of the child. With this information, health care providers could provide the help and support these families might need. This support might exist of trying to lower the fear of hypoglycemia in parents. Research has shown that interventions including blood glucose awareness training and cognitive behavioral therapy can reduce these levels of fear [78]. Furthermore, health care providers could provide help in order to lower parenting stress by, for example, a mindfulness-based stress reduction [79] or a specific program developed for parents with a child with T1DM called *DELFIN* (Das ELterntraining fur Eltern vor kINdern met Diabetes Typ 1) [80], which both have shown promising results [79, 80]. Children could be supported by lowering a possible needle-phobia by, for example, using stress-reducing medical devices which is a new, effective cognitive therapy for needle phobia with promising results [81].

Furthermore, it is also important to gather information about levels of stress of both parents, as fathers and mothers seem to perceive the amount of pediatric parenting stress differently (**Chapter 6**). It appears that the involvement of fathers in their child's treatment is associated with better family, maternal and marital family outcomes [42]. Therefore, both parents should be included in the diabetes-treatment of their child and we recommend to assess the Pediatric Inventory for Parents [82] periodically in both the father and the mother of the child. This would enable health care providers to determine whether one or both parents are in need for support (like participating in the *DELFIN* program [80]).

Finally, a secure attachment relationship could reduce child behavior problems and improve the diabetes-specific QoL in families who experience high levels of pediatric parenting stress (**Chapter 7**). Therefore, it might be important for these families to provide help and support to develop a secure attachment relationship by using VIG to improve parental insensitivity mentioned earlier in this chapter. Furthermore, besides discussing only the obvious child outcomes like the HbA_{1c} level and QoL, health care providers should also regularly discuss the experienced amount of pediatric parenting stress, and child behavior problems with the parents. Although some hospitals already offer 'carrousel-

appointments' for their patients, in which all outcomes and possible problems are discussed with several disciplines on a regular basis, this should be implemented as a standard in the care of children with T1DM and their parents.

Concluding remarks

The results described in the present thesis support the notion that T1DM is a family disease: it not only affects the children, but also their parents. The quality of parent-child interactions and pediatric parenting stress are related with central child outcomes (HbA_{1c} level and QoL). A causal relationship still remains unclear. Therefore, future research is needed to examine, for example, whether pediatric parenting stress causes or is caused by a low QoL of the child or whether other common denominators (such as poverty or a bad marriage) play a role. With this information, health care providers can specifically focus their treatments on the parent or child in order to improve the QoL of the child. Also, because research on the quality of attachment relationship in young children with T1DM is scarce, future research should focus on this aspect, as a secure attachment relationship could reduce child behavior problems and improve the diabetes-specific QoL in families who experience high levels of pediatric parenting stress. Health care providers should, therefore, focus on the family as a whole and not only on the glycemic control and psychosocial outcomes of the child, as family factors like parent-child interactions, parenting stress, and attachment are related or have an influence on one or more of these child outcomes.

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About the author

ABOUT THE AUTHOR

Anke Nieuwesteeg was born on July 30, 1985 in Someren, the Netherlands. She completed her pre-university education at the Varendonck College, Asten, in 2003. From 2003 to 2007 she studied at Radboud University Nijmegen where she obtained the degrees Bachelor of Science (BSc) and Master of Science (MSc) in behavioral sciences (learning and developmental problems – specializing in sensory impairments). Thereafter, she started working as a behavioral scientist and later as a coordinator of care at Kempenheaghe, Heeze, a center for adults with epilepsy and intellectual disabilities. In 2010 she started her PhD research at Tilburg University, supervised by Prof. dr. Hedwig van Bakel, Prof. dr. Frans Pouwer and dr. Esther Hartman. Her research focused on the quality of parent-child interaction and pediatric parenting stress in families with a young child with type 1 diabetes mellitus. Currently, she is working for the the Medical Ethics Review Committee at Màxima Medical Center.